CASE REPORT

Unusual organism causing pyocystis in an immunosuppressed haemodialysis patient

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SUMMARY

A 68-year-old man with end-stage renal disease and a history of orthotopic cardiac transplantation on chronic immune suppression therapy presented to the emergency department with fever and purulent urethral discharge. He was diagnosed with pyocystis (bladder abscess). Culture of the urethral discharge showed *Trueperella bernardiae*. The patient improved after 3 days of bladder irrigation with normal saline and gentamicin. This case demonstrates that bladder abscess is a potential source of infection in an oliguric patient. This is the first report to describe the opportunistic pathogen, *T. bernardiae*, as the causative agent in pyocystis.

BACKGROUND

Infection is a leading cause of death in renal dialysis patients.¹ Given the increased prevalence of dialysis worldwide, clinician awareness of pyocystis is important to prevent complications and reduce mortality.^{1–2} Rapid identification of pyocystis is essential as bladder irrigation is necessary to sufficiently eradicate infection. Patients on chronic immunosuppression are at risk of infection from opportunistic organisms. This case raises awareness of pyocystis and *Trueperella bernardiae*, a recently characterised but poorly understood opportunistic pathogen that can cause bladder abscess.

CASE PRESENTATION

A 68-year-old man with a history of orthotopic cardiac transplantation, end-stage renal disease (ESRD) on haemodialysis for 8 years and stroke, presented to the hospital with fever and penile discharge. The patient's caretaker noted the presence of occasional blood clots in the patient's urine for several weeks. Four days prior to admission, the patient became increasingly lethargic and developed large amounts of purulent penile discharge. He developed a fever of 102.7°F and was brought in for evaluation. He denied chest pain, shortness of breath, flank pain, abdominal discomfort and urinary burning. His frequency of urination—a small volume of urine usually once a day—did not change. His medications included cyclosporine and mycophenolate mofetil for immunosuppressive therapy related to cardiac transplantation performed 12 years previously for idiopathic cardiomyopathy. His renal disease developed secondary to taking these medications. He had no history of opportunistic infections.

On admission, the patient had a heart rate of 99 bpm, temperature of 98.2°F, blood pressure of 120/76 and respiratory rate of 20. His general

appearance was of a thin, lethargic and ill-appearing man. He was oriented to his name but not the hospital location or date. Abdominal examination was unremarkable. His extremities were warm and well perfused. Genitourinary examination was significant for white-yellow purulent fluid draining from the urethral meatus. On digital rectal examination, the prostate was firm, smooth, non-enlarged and non-tender. A Foley catheter was placed and drained bloody purulent material.

INVESTIGATIONS

A complete blood count showed a white cell count of $5.9 \times 10^9/L$ with 72.4% neutrophils. Blood urea nitrogen was 21 mg/dL and serum creatinine was 4.88 mg/dL. Chest X-ray was normal. CT of the brain showed no acute changes. The patient's cyclosporine level was in the therapeutic range during admission (200–400 ng/mL).

An ultrasound revealed atrophic kidneys and echogenic debris within the bladder, without evidence of flow (figure 1). Culture of the discharge revealed >10 000 colonies of *T. bernardiae*. Four sets of blood cultures showed no growth.

DIFFERENTIAL DIAGNOSIS

Purulent penile discharge in the setting of lethargy and fever points to an infection involving the lower urinary system. The differential diagnosis included urethritis, cystitis and prostatitis. Pyocystis was considered the most likely diagnosis given the large volume of purulent discharge, history of oliguria and echogenic debris filling the bladder on ultrasound imaging.

TREATMENT

The patient was started on intravenous ceftriaxone once daily and bladder irrigation with a solution of

Figure 1 Ultrasound image of the urinary bladder (sagittal view) displaying large amounts of echogenic material within the bladder.

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gentamicin and normal saline. The irrigations were performed three times a day. On hospital day 4, the patient's symptoms improved dramatically. He was transitioned from ceftriaxone to a 5-day course of oral cefuroxime. Once weekly irrigation with normal saline was arranged.

OUTCOME AND FOLLOW-UP

The patient rapidly improved after daily bladder irrigations with normal saline and gentamicin. His urine became clear after 5 days of daily irrigation. He was discharged home with nursing services arranged to catheterise and irrigate his bladder once a week with normal saline for 4 weeks. Outpatient follow-up with a urologist for haematuria and malignancy work up was arranged.

DISCUSSION

Pyocystis is an often overlooked source of infection among anuric patients.^{3 4} More commonly it has been reported in patients with supravescicular urinary diversion and a retained bladder with an incidence ranging from 10% to as high as 67%.^{3 5} In the ESRD dialysis-dependent population, a lack of diuresis leads to defunctionalisation of the bladder and sloughing of the urothelial wall.^{3 5 6} Fluid stasis further promotes bacterial colonisation. These factors generate a microenvironment in which the bladder wall is predisposed to abscess formation.^{5 6} Susceptibility is further increased by comorbid conditions including diabetes or immunosuppression.⁴ Unavailability or delays of urine specimens that can be provided for microbiological testing as well as altered immune responses in dialysis patients may lead to a delay in diagnosing pyocystis. Untreated patients may develop sepsis and/or extension of the abscess to adjacent tissues.⁷

The typical presentation of pyocystis involves oliguric patients who present with signs of infection such as fever and lethargy. Local symptoms may be present and can include dysuria, change in urinary frequency, haematuria, pelvic pain and purulent discharge. Organisms causing pyocystis in immunocompetent patients are typical urinary pathogens including *Escherichia coli*, *Proteus* and *Klebsiella* spp.^{3 8} Imaging with ultrasound is often helpful as it can reveal localised abscesses within the bladder wall.

Identification of pyocystis is necessary to adequately treat this infection.^{4 8} In contrast to systemic antibiotic therapy for typical urinary tract infections (UTIs), standard treatment for pyocystis includes bladder irrigation.⁹ Bladder washout, often with intravesical antibiotic administration, is performed because systemic antibiotics have poor penetration into the defunctionalised bladder.^{5 8} Regular irrigation also functions to clear the bladder of purulent material.^{4 5 8 10} Bactericidal solutions associated with a positive response include gentamicin, nitrofurantoin, silver nitrate, chlorhexidine and acetic acid.^{4 11} Coverage with intravenous antibiotics is reasonable to treat and prevent concomitant bacteraemia and pyelonephritis. Two to 4 weeks of therapy has been anecdotally recommended.⁵ In selected cases, particularly in patients with prior urinary diversion, interventions such as cystostomy or cystectomy may be required to eradicate resistant or recurrent infection.⁹

This is the first case to report pyocystis formation from *T. bernardiae*. This organism was first described by the US Centers for Disease Control as an opportunistic pathogen belonging to the coryneform group 2 bacteria genus.¹² It was reclassified into the *Arcanobacterium* genus in 1997 and finally into the newly described *Trueperella* genus in 2011.^{13 14} *T. bernardiae* is a predominantly facultative anaerobic Gram-positive

coccobacillus that is non-motile and non-sporulating. This organism is catalase negative and does not reduce nitrites.¹²

The pathobiology of *T. bernardiae* is not well understood. Case reports describe this organism as the causative agent in UTIs involving patients with diabetes or prior urinary diversion surgeries.^{13–15} In this setting, the organism was capable of causing perineal abscess, bacteraemia and even thoracic empyema.^{16–18} *T. bernardiae* has also been implicated in bone and soft tissue infections.^{15 19–21} One report describes this organism as the agent in septic arthritis of the hip in a patient receiving immunotherapy for systemic lupus.¹⁵ Prosthetic joint infection of an elderly man was also reported.¹⁹ These findings suggest that *T. bernardiae* is an opportunistic pathogen capable of causing both localised abscesses and systemic infections.

There is no standard treatment for *T. bernardiae*. Antibiotic therapy varies depending on location, severity and culture-derived susceptibilities. The patient in this case demonstrated sufficient response to intravenous ceftriaxone followed by oral cefuroxime and standard pyocystis treatment using bladder irrigation. Other antibiotics associated with a positive response include rifampin with oxifloxacin, ceftriaxone and clindamycin.^{15 21} Efforts to understand the pathogenicity of *T. bernardiae* and the patterns of human infection with which it is associated are necessary to optimise treatment.

Learning points

- ▶ Pyocystis (bladder abscess) is an important and easily overlooked source of infection in dialysis patients.
- ▶ Bladder irrigation with normal saline and antibiotics is central to the treatment of pyocystis.
- ▶ Chronically immunosuppressed, oliguric patients may be at risk of pyocystis infections from atypical organisms.
- ▶ *Trueperella bernardiae* is an opportunistic anaerobic Gram-positive organism capable of causing pyocystis.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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