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CASE REPORT

Burkholderia cenocepacia urosepsis with a perinephric haematoma in an immunocompetent patient with ureteral calculus obstruction

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Introduction

Sepsis caused by acute ureteral calculus obstruction is life threatening, requiring immediate renal decompression and empiric antibiotic therapy. Delaying drainage of an obstructed and infected kidney can cause sepsis, acute renal failure and death. *Burkholderia cenocepacia* is a Gram-negative biofilm-forming bacterium which is often multidrug resistant. It is an opportunistic pathogen in patients with cystic fibrosis or immunosuppression. To the best of the authors' knowledge, there have been no previous reports of urosepsis caused by this bacterium. This article describes the case of an immunocompetent man who developed *B. cenocepacia* urosepsis superimposed on calculus ureteric obstruction.

Case report

A 48-year-old man presented to the emergency department with right flank pain and leucocytosis (white cell count 15.3x10^9/litre). He had recently had extracorporeal shockwave lithotripsy for right-sided renal stones and the treatment was uneventful. Computed tomography of the kidneys, ureters and bladder showed a 7 mm calculus in the proximal third of the right ureter with moderate hydroureteronephrosis. The patient declined a ureteric stent and was discharged home with analgesia and oral co-amoxiclav 625 mg three times daily for 5 days.

He re-presented 3 weeks later with right-sided flank pain, fever and altered sensorium. He was tachycardic, hypotensive and had raised inflammatory markers (white cell count 19.3x10^9/litre), C-reactive protein (313 mg/dl) and lactate (3.8 mmol/litre) levels, so urosepsis was diagnosed. He was HIV negative, and had not received treatment with corticosteroids or biologicals which could have led him to be immunocompromised.

He was resuscitated with intravenous fluids and started on intravenous co-amoxiclav and amikacin. A repeat computed tomography of the kidneys, ureters and bladder scan revealed a right proximal ureteric calculus (7 mm), moderate right-sided hydroureteronephrosis and perinephric stranding (Figure 1a). He underwent a nephrostomy under ultrasound guidance with an 8 French locking pigtail catheter (Fleixma pigtail catheter, Boston Scientific, Massachusetts, USA); there were no procedural complications. Three sets of blood cultures and two urine cultures grew *Burkholderia cenocepacia*. Bacterial identification was by matrix-assisted laser desorption/ionisation – time of flight analyser (MALDI-TOF) mass spectrometry in the local laboratory, later confirmed by the same method at the reference laboratory. The antibiotic sensitivity report from the reference laboratory were based on Clinical and Laboratory Standard Institute guidelines, adjusted to breakpoints in the reference laboratory antimicrobial sensitivity method. The minimum inhibitory concentrations and breakpoint of the blood culture isolate reported by the reference laboratory are in Table 1. Antibacterials were changed to ceftazidime 2 g intravenous three times daily.

Following a nephrostogram 4 days later, the patient became haemodynamically unstable. A repeat computed tomography of the kidneys, ureters and bladder scan showed a perinephric and subcapsular haematoma in the anterior aspect of the right kidney suggestive of recent haemorrhage. A renal angiogram done immediately afterwards showed active arterial bleeding into the haematoma, which expanded during the 40 minutes between computed tomography scans (Figure 1b and c). Therefore, emergency selective renal embolisation was performed (Figure 1d). During embolisation, abnormal vascular friability led to dissection and occlusion of the bleeding vessels, extending to the main interpolar artery. To treat the dissection, he had an angioplasty with a 4 mm balloon (Cook Medical, Indiana, USA) which resulted in loss of approximately 20% perfusion of the right kidney.

Case report (continued)

The patient completed 5 days of ceftazidime and then 7 days of intravenous meropenem as he developed a skin rash with ceftazidime. He went home with a nephrostomy in situ.

Ten days post-discharge the patient returned for antegrade right ureteric stenting and nephrostomy removal. Two weeks later he underwent right flexible ureteroscopy, laser fragmentation and change of stent. He was symptom free after that, and his renal function tests are normal.

Table 1. Minimum inhibitory concentrations and breakpoints of the blood culture isolate

Antibiotic	Minimum inhibitory concentration (mg/litre)	Sensitive (S), intermediate (I) or resistant (R)	Breakpoint (mg/litre)
Amikacin	>64	R	8 and 16
Gentamicin	>32	R	4
Tobramycin	>32	R	4
Aztreonam	2	I	1 and 16
Ceftazidime	1	S	8
Imipenem	8	I	4 and 8
Meropenem	2	S	2 and 8
Piperacillin/tazobactam	≤1	S	16
Ciprofloxacin	>8	R	0.5 and 1
Minocycline	8	R	2 and 4

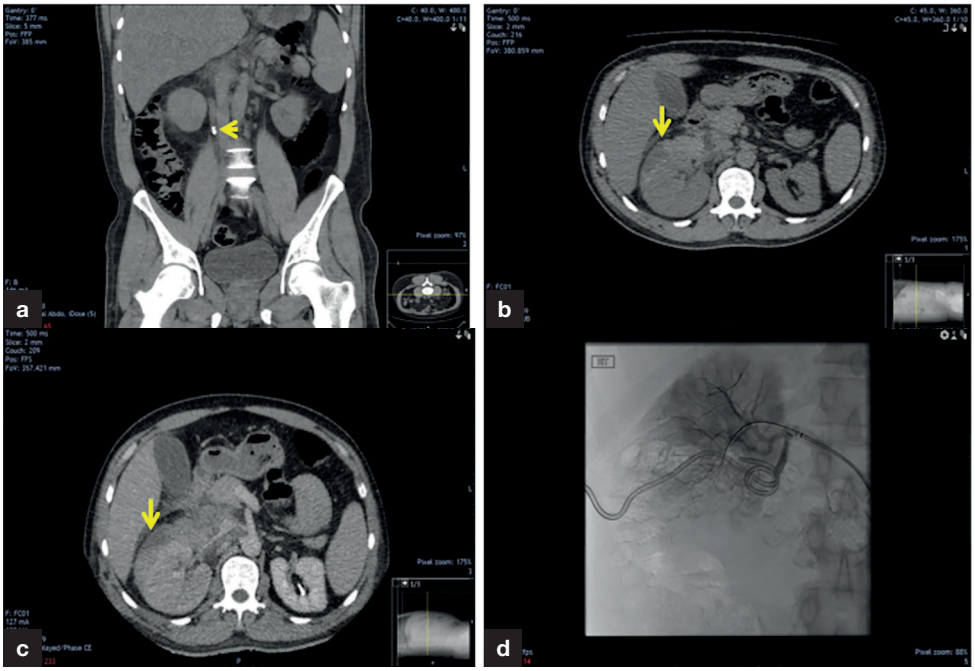

Figure 1. a. A computed tomography scan showing hydroureteronephrosis secondary to an upper third ureteric stone (arrow). Perinephric stranding present suggestive of inflammation. b. A non-contrast computed tomography scan showing the haematoma (arrow). c. A computed tomography renal angiogram showing an increase in the size of the haematoma over 40 minutes (arrow). d. A selective right renal angiogram in progress. The lower pole of the kidney is poorly perfused.

Discussion

Ureteral obstruction in septic patients requires immediate decompression and antibiotics. Delay can cause renal failure or death.

B. cenocepacia, a member of the *B. cepacia* complex, comprises nine genomovars. These are nosocomial pathogens, particularly in patients receiving broad-spectrum antibacterials (Lipuma, 2005). These non-aeruginosa opportunistic pseudomonads are resistant to multiple antibiotics including aminoglycosides, penicillins, third-generation cephalosporins and polymyxin B. *B. cenocepacia* bacteraemia is seen predominantly in immunocompromised patients and those with cystic fibrosis in whom it is a chronic coloniser (Lipuma, 2005). *B. cenocepacia* has spread epidemically within populations of people with cystic fibrosis in Canada and Europe (Kenna et al, 2017). Although infections are reported in intensive care units and pregnancy, it rarely causes urinary infections in immunocompetent patients (Katsiari et al, 2012; Ishtiaq et al, 2017; Gopalratnam et al, 2018). A UK-wide analysis of *Burkholderia* in patients who did not have cystic fibrosis showed only 30 isolates from 25 patients over 2 years (Kenna et al, 2017).

Patients with urinary calculi often have positive stone cultures. Studies sequencing microbiota of urolithiasis show that bacteria aggregate selectively to crystals, that their presence is associated with crystal clumping, and that they stimulate protein incorporation into the stone matrix (Schwaderer and Wolfe, 2017). *B. cenocepacia* forms biofilms, so it is possible that by creating a biofilm on stones, bacteria evade phagocytic neutrophils and cause localised inflammation (Murphy and Caraher, 2015).

This patient was not immunocompromised. Apart from prior extracorporeal shockwave lithotripsy, he did not have any history of medical treatment or invasive procedures and the source of this infection remains conjectural. However, his treatment with broad-spectrum antibiotics and delayed renal decompression because he refused a ureteric stent may have contributed to the susceptibility to this organism.

Subcapsular and perinephric haematomas may occur after extracorporeal shockwave lithotripsy and ureteroscopic lithotripsy (Knapp et al, 1988; Kozminski et al, 2015). However, subcapsular and perinephric haematoma associated with *B. cenocepacia* urosepsis has not been described previously. *B. cenocepacia* produces toxins, including surface-active haemolysins, contributing to a profound inflammatory response (Hutchison et al, 1998). Conceivably, these toxins, combined with a host inflammatory response, caused vascular degradation and friability of intrarenal vessels, leading to secondary haemorrhage. The secondary haemorrhage and subcapsular and perinephric haematoma may have been precipitated by the nephrostogram, aggravating trauma caused by the earlier nephrostomy. Vascular friability may also account for the arterial dissection during angioplasty.

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Learning points

- There are no prior reports of urosepsis secondary to *Burkholderia cenocepacia.*
- *B. cenocepacia* is a Gram-negative highly multi-drug resistant biofilm-forming organism, that rarely infects immunocompetent patients.
- Sepsis caused by acute ureteral obstruction requires urgent decompression of the obstructed kidney and treatment with antimicrobials.
- *B. cenocepacia* haemolysins, biofilm-enabled evasion of neutrophilic phagocytosis, and a strong host response could have led to intrarenal vascular complications contributing to the complexity of managing this patient.
- Emerging infections and their diverse manifestations should be kept in mind.

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