



Case report

Out of this world: *Elizabethkingia miricola* complicated urinary tract infection in a patient with associated pubic symphysis osteomyelitis and pyomyositis

Karim Badawi^{a,*}, Seth Deskins^b, Kristen Catherman^a, Allison Lastinger^{b,c}

^a Department of Medicine, West Virginia University School of Medicine, Morgantown, WV, the United States of America

^b Department of Internal Medicine/Pediatrics, West Virginia University School of Medicine, Morgantown, WV, the United States of America

^c Division of Infectious Diseases, West Virginia University School of Medicine, Morgantown, WV, the United States of America

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ABSTRACT

Elizabethkingia miricola (*E. miricola*) is a gram-negative rod initially isolated from condensation at the Russian Mir space station. In the literature, there are few cases of human isolates that have been identified, with only one prior case of *E. miricola* urinary tract infection (UTI). Here we report a case of a patient with a chronic suprapubic catheter that was found to have *E. miricola* UTI with fistulization between the bladder and pubic symphysis, leading to osteomyelitis and surrounding pyomyositis. He was placed on Tigecycline based on susceptibility profile, underwent bilateral nephrostomy tube placement and discharged home with close outpatient follow-up. With the increasing use of novel detection methods, accurate identification and antimicrobial susceptibility testing is necessary for this multidrug resistant organism and others like it.

Introduction

E. miricola is a non-fermenting, gram-negative, strict aerobic, oxidase positive rod first isolated from condensation in the Russian space station Mir in 2003 [1]. Very few cases of human inoculation have been identified, with only one case of *E. miricola* urinary tract infection being reported [2]. However, with the use of matrix-associated laser desorption ionization time-of-flight mass spectrometry (MALDI-TOF-MS) on the rise, the identification of non-fermenting pathogens has led to increased detection and understanding of the clinical significance of rare multi-drug resistant microorganisms. Here we present a case of *E. miricola* UTI with associated pubic symphysis osteomyelitis and left lower extremity pyomyositis.

Case report

A 67-year-old male with complex past medical history of hypertension, hyperlipidemia, obstructive sleep apnea, stage 3 chronic kidney disease with baseline creatinine around 1.10, bilateral lower extremity neuritis/plexitis and prostate cancer status post radiation (2012) which was complicated by urethral stricture requiring placement of a suprapubic catheter. He presented with a one-week history of progressively

worsening left thigh pain, erythema, swelling, and tenderness. He was recently diagnosed with prostatitis which was being managed by infectious disease. He had recently been treated with a six week course of Ciprofloxacin before transitioning to Trimethoprim-Sulfamethoxazole due to lack of symptomatic improvement. He was also noted to have grown *E. miricola* on urine cultures one and two weeks prior to presentation. Of note, around this same time he was undergoing evaluation at the neuromuscular clinic for weakness and severe proximal lower extremity pain with abnormal NCS/EMG indicating prolonged bilateral peroneal F wave latencies with abnormal right peroneal nerve conduction studies without electrophysiological evidence of an underlying myopathy. For this he was started on a prednisone 50 mg daily, which he remained on due to exacerbation of pain with attempted tapering of the dose.

Physical exam at admission was remarkable for abdominal tenderness, intact suprapubic catheter draining clear urine, and left thigh erythema, swelling, warmth with tenderness to palpation. No discernable abscess was palpated. Laboratory workup revealed leukocytosis of $11.9 \times 10^3/\mu\text{L}$ with a $10.12 \times 10^3/\mu\text{L}$ neutrophilic predominance, C-reactive protein (CRP) of 77.7, erythrocyte sedimentation rate of 67, and a nonoliguric acute kidney injury with a creatinine of 1.49 mg/dL. Urine and blood cultures were obtained on admission and showed no growth.

* Corresponding author.

E-mail address: kabadawi@hsc.wvu.edu (K. Badawi).

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Computerized tomography (CT) bilateral lower extremities with IV contrast revealed new peripherally enhancing, irregular, loculated, fluid density collections seen at the symphysis pubis with osseous erosions of the bilateral symphyseal bodies compatible with the diagnosis of osteomyelitis, with the fluid densities extending into bilateral pectineus, obturator, left adductor longus, and left adductor magnus muscles consistent with pyomyositis (Fig. 1).

General surgery was consulted by the emergency department and did not recommend any surgical intervention. He was admitted to the medicine service for further workup and intravenous antibiotics. Based on *E. miricola* sensitivity profile from previous urine cultures prior to presentation, he initially started on empiric therapy of Gentamicin, Vancomycin, and Flagyl. Infectious disease was consulted and recommended Aztreonam, Tigecycline, and one time dose of Amikacin; meanwhile, *E. miricola* urine cultures from one week prior to presentation were sent to Associated Regional and University Pathologists for a susceptibility panel, which revealed the isolate to be minocycline susceptible but Aztreonam resistant (Table 1). Aztreonam was discontinued with the continuation of Tigecycline for six weeks due to the presence of osteomyelitis. He had rapid improvement in left medial thigh swelling and erythema. Due to concern for colonization, the patient's suprapubic catheter was exchanged with repeat urine cultures from the new catheter also without growth during the admission.

Urology was consulted and recommended CT cystogram with IV contrast to assess for possible urinary leak. This study revealed fistulization between the suprapubic catheter tract/bladder and the loculated air and fluid collection along the anterior pubic symphysis (Fig. 2). The patient subsequently underwent bilateral percutaneous nephroureteral tube placement for urinary diversion with the ultimate goal for the fistula to close.

Neurology was consulted while inpatient for further management/plan regarding his Prednisone. They recommended that he have intravenous immunoglobulin (IVIG) infusions 2 g over three days, with plan to taper the Prednisone by 10 mg every three days following. Along with this, he was continued on Trimethoprim-Sulfamethoxazole for pneumocystis pneumonia prophylaxis. Outpatient follow-up was arranged with genitourinary reconstructive surgery, with plans for possible supratriangular cystectomy and ileal conduit urinary diversion.

Discussion

E. miricola is an infectious pathogen with increased incidence of isolation in humans. A case series analyzing the risk factors for

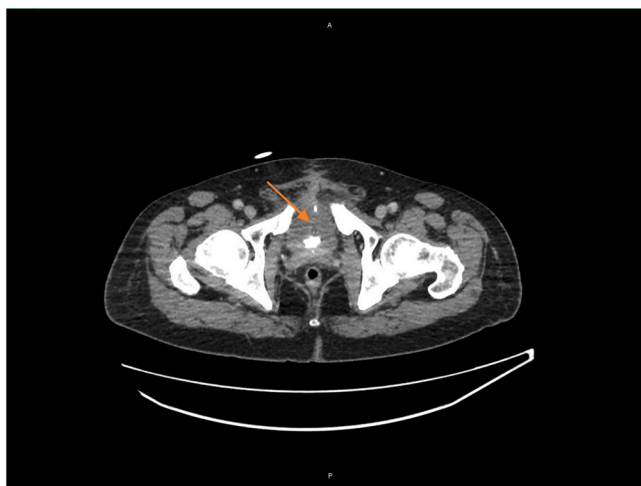


Fig. 1. CT bilateral lower extremity with IV contrast revealed fluid collections and osseous erosions at pubis symphysis concerning for osteomyelitis, as well as bilateral extension into surrounding musculature concerning for pyomyositis.

Table 1

The drug susceptibility of *Elizabethkingia miricola* isolated from patient.

Antibiotic	MIC
Amikacin	≥ 64 Resist
Gentamicin	≥ 16 Resist
Cefepime	≥ 64 Resist
Minocycline	1 Suscept
Imipenem	≥ 32 Resist
Meropenem	≥ 16 Resist
Ciprofloxacin	≥ 8 Resist
Trimethoprim/Sulfamethoxazole	≥ 8/152 Resist
Piperacillin/Tazobactam	≥ 128/4 Resist
Aztreonam	≥ 64 Resist
Ceftazidime	≥ 32 Resist
Levofloxacin	4 Intermed
Ticarcillin/Clavulanate	≥ 256/2 Resist
Tobramycin	128 Resist

MIC = minimum inhibitory concentration.

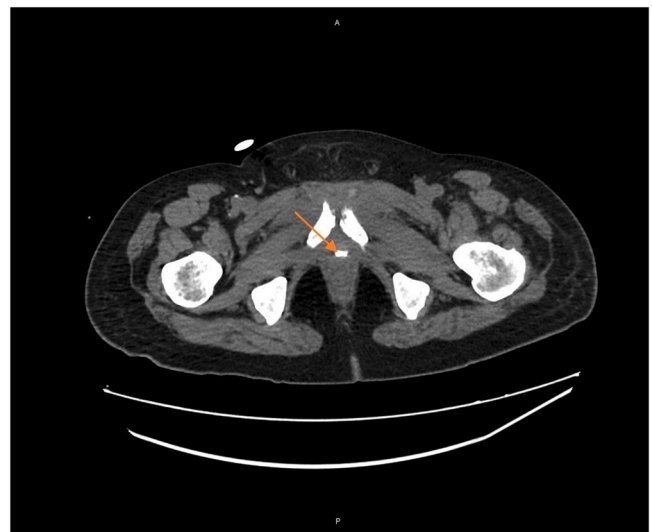


Fig. 2. CT cystogram with suspected extravasation indicating fistulization between the suprapubic catheter tract/bladder and the loculated air and fluid collection along the anterior pubic symphysis.

E. miricola was conducted in South Korea between 2016 and 2017 and found that incidence has increased by 432.1 % as compared to 2009–2015, with factors such as hospital environment and mechanical ventilation being associated with the acquisition of *Elizabethkingia* species [4]. The first identified case was in 2008 in a patient with stage IV mantle cell lymphoma who was intubated and mechanically ventilated due to prolonged neutropenia following salvage chemotherapy leading to diffuse pulmonary infiltrates. This patient was noted to develop a new fever on day 17 post-ICU admission, respiratory cultures grew an organism that was later identified as *E. miricola*. Subsequent blood cultures were also found to grow this organism. The isolate was susceptible to levofloxacin and tigecycline, with fever resolving following 48 h of tigecycline. Levofloxacin was added to complete two weeks of therapy [3]. Only one case of *E. miricola* UTI has been identified prior to this case. This study reported isolation of *E. miricola* in a 25-year-old female presenting with a one-month history of increased bowel frequency, oliguria, fever, and abdominal pain. That isolate was sensitive to gentamicin, ceftriaxone, aztreonam, piperacillin-tazobactam and imipenem, with resistance noted to ampicillin, ciprofloxacin, levofloxacin, vancomycin and colistin. The patient was started on piperacillin-tazobactam and responded well with resolution of symptoms, and sterile urine cultures upon two week follow-up [2]. In our literature review, we could find no documented cases of osteomyelitis due to *E. miricola*. While pelvic bone biopsy was not pursued for our patient, we felt that given multiple urine

cultures growing multi-drug resistant *E. miricola* in the setting of a direct conduit from the bladder to the pelvis, this possibly represents the first case of osteomyelitis from this microbe.

Antibacterial selection is based on the susceptibility of the microbe. For all multidrug resistant organisms, it is imperative we collect sensitivity data to guide appropriate initial antibiotic selection for future infections. Our isolate was solely sensitive to Minocycline (Table 1), and it should be noted that our patient continued to improve clinically and objectively, as noted by resolution of symptoms, leukocytosis and down trending C-reactive protein with recommended antibiotics based on susceptibility profile, as well as in the cases listed above [2,3]. Tigecycline is not typically used to treat urinary tract infections due to its poor urinary penetration, but in this case, it was felt to be the best drug given the susceptibility and the direct communication between the bladder and the pubic symphysis bone.

In summary, we report a case of *E. miricola* UTI with associated pubic symphysis osteomyelitis and pyomyositis. While this pathogen has been increasingly isolated recently, it remains an exceedingly rare cause of UTI, with only one prior report in literature, and only a handful of other cases causing profound sepsis. Susceptibility-driven therapy is critical for successful treatment of this rare microorganism.

Ethical approval

Not applicable.

Consent

Informed consent not obtained due to lack of personal or identifiable information in this report.

CRedit authorship contribution statement

KB and SD designed the case report and drafted the manuscript. KB, SD, KC, and AL diagnosed and treated the patient. AL advised on the treatment of elizabethkingia in this patient. KB, SD, KC, and AL revised the manuscript. All authors have approved this manuscript for submission.

Competing Interest

We have no conflict of interest to declare.

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