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Case report

A capricious case of *Staphylococcus caprae* thoracic osteomyelitis

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ABSTRACT

Staphylococcus caprae (*S. caprae*) is a gram positive, coagulase-negative *Staphylococci* (CoNS) that occurs as a commensal pathogen on the human skin. It recently has been recognized in causing nosocomial infections involving the bloodstream, urinary tract, heart, bone, and joints, particularly in immunosuppressed patients or individuals with prosthetic devices. Previously, *S. caprae* was underreported as it was difficult to identify in the clinical microbiology laboratory; however, due to advances in molecular identification methods and matrix-assisted laser desorption ionization time-of-flight mass spectrometry (MALDI-TOF MS), more clinical cases are being identified in human isolates and appropriately treated. *S. caprae* osteoarticular infections are usually associated with polymicrobial infections and presence of orthopedic prostheses in immunocompromised adults. This pathogen has an even rarer presentation of bone and joint infections (BJIs) in immunocompetent individuals without orthopedic devices.

Our case is of a 65-year-old immunocompetent male with diet-controlled diabetes mellitus type 2 and end-stage renal disease (ESRD) on hemodialysis who presented with worsening mid-thoracic pain after a ground-level fall and was diagnosed with biopsy-proven *S. caprae* thoracic discitis/osteomyelitis, associated with recurrent catheter-related bloodstream infection (CRBSI). It illustrates the importance of recognizing *S. caprae* as an emerging human pathogen, even in immunocompetent individuals without orthopedic hardware, requiring prompt targeted treatment of native BJIs to prevent unfavorable outcomes.

Introduction

Coagulase-negative *Staphylococcus* (CoNS) species are generally considered low risk to healthy humans and in some situations protect against invasion by other pathogenic or harmful bacteria. *Staphylococcus caprae* is a commensal CoNS species first documented in 1983 as a cause of mastitis in goats after being isolated from goat milk [1–4]. In humans, *S. caprae* has been known to colonize skin, nose, and nails. This organism has been reported in several cases as a pathogen, causing both community-acquired and hospital-acquired infections in humans including peritonitis, meningitis, bacteremia, urinary tract infections, infective endocarditis, prosthetic joint infections, and osteomyelitis.

S. caprae has been implicated in bone and joint infections (BJIs) usually after orthopedic operations; however, recently there have been very rare cases of this pathogen occurring in native BJIs without orthopedic prostheses. Risk factors for developing *S. caprae* infections include physical contact with goats or sheep (farmers, sheep breeders, and those who had been bitten by goats), diabetes mellitus, chronic renal failure, immunosuppression, open traumatic fractures, and obesity [5]. Recent advancements in molecular techniques have improved identification of *S. caprae* strains due to MALDI-TOF MS [1,3]. *S. caprae* is an underrepresented commensal pathogen that should be included in the differential for BJIs, even for individuals without orthopedic hardware.

Most published literature on *S. caprae* bone and joint infections involve orthopedic devices, with only a few studies available that discuss cases without retained hardware. In this case report, we discuss a patient that developed *S. caprae* native vertebral osteomyelitis of the thoracic spine complicated by CRBSI.

Case presentation

A 65-year-old male with chronic low back pain presented to the emergency room with a two-week history of right rib pain after a ground

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level fall at home. His medical history was significant for end-stage renal disease (ESRD) requiring hemodialysis (HD) via his left internal jugular tunneled line, diabetes mellitus type 2, hypertension, hypothyroidism, pulmonary sarcoidosis on no current therapy, and stable ascending aortic aneurysm diagnosed five years prior. He described his rib pain as sharp and radiating down his back but denied any symptoms of fevers or chills, loss of bowel or bladder continence, or recent antibiotic use.

On admission, he was afebrile with a temperature of 98.1°F and blood pressure 151/75 and lab tests showed a white blood cell (WBC) count of 7570/mm^3, hemoglobin of 11.1 mg/dL, platelet count of 415,000/mm^3, creatinine of 4.90 mg/dL, erythrocyte sedimentation rate (ESR) of 75 mm/hr, c-reactive protein (CRP) of 25.8 mg/L, and an unremarkable urine drug screen and urinalysis. Rib x-ray and computed tomography (CT) cervical spine showed no acute fracture or subluxation. He underwent further imaging with CT chest, abdomen, and pelvis revealing new endplate changes at the level of T7-T8 with paravertebral soft tissue thickening concerning for early discitis/osteomyelitis and cirrhotic liver with splenomegaly and ascending aortic aneurysm 4.4 cm at the level of pulmonary artery. Neurosurgery service was contacted for further evaluation and recommended the patient undergo a CT-guided bone biopsy for work-up and to defer antibiotics to increase microbiological yield, given the patient was hemodynamically stable and without leukocytosis. The following day, magnetic resonance imaging (MRI) of thoracic and lumbar spine confirmed T7-T8 discitis/osteomyelitis with no canal stenosis or compression, multilevel spondylosis and degenerative disease most prominently of L3-S1 (Figure 1). Blood cultures from peripheral venous sites and the HD line were collected on admission and he underwent CT-guided bone biopsy on day three of hospitalization. Patient's thoracic bone biopsy and blood cultures from his HD line grew *Staphylococcus caprae (S. caprae)*. Empiric IV vancomycin was started.

Infectious diseases (ID) were consulted on day 5 for antibiotic recommendations. Further history was obtained by patient interview and medical records review. Three months prior to current presentation, the patient had been admitted for a complaint of worsening lumbar back and abdominal pain. Abdominal work-up with ultrasound and MRI abdomen confirmed cholelithiasis without evidence of acute cholecystitis and dilated common bile duct without choledocholithiasis. MRI of thoracic and lumbar spine was negative for thoracic discitis/osteomyelitis and consistent with multilevel degenerative changes of his lumbar spine (Figure 2). Lab tests showed a mildly elevated ESR of 31 mm/hr and CRP of 31.4 mg/L. Blood cultures from peripheral venous blood were obtained in the ED due to concern for infection and his nephrologist drew one set of blood cultures from his HD line. *S. caprae* grew in one of two bottles from his HD line. The CoNS species was initially suspected to be a skin contaminant and long-term antibiotics were held after receiving an empiric dose of IV vancomycin. Repeat blood cultures were obtained from peripheral venous blood and his HD line; *S. caprae* grew again from his HD line culture while his peripheral blood culture had no growth. Therefore, his tunneled right internal jugular line was removed and after a 72-hour line holiday, a new tunneled left internal jugular line was placed. He completed a seven-day course treatment with vancomycin and de-escalated to cefazolin while inpatient. The patient disclosed that he had no recent travel outside of Georgia or South Carolina for several years; however, he spent thirteen months in South Korea when he was on military active duty. He had exposure to a one-year-old dog in his neighborhood but denies any recent scratches or bites. He had no contact history with sheep, goats, cattle, chickens, or raw milk. He also reported that he had no prior history of malignancy, HIV/AIDS, or recent use of high dose corticosteroids or biological agents for his sarcoidosis.

Final antimicrobial susceptibility report was reviewed (Table 1). ID recommended to continue renally dosed IV vancomycin and to pursue another line holiday. After achieving clearance of his bacteremia, a new vascular access was placed, and he was discharged to complete a six-week course of IV vancomycin with his outpatient HD sessions. After completing five weeks of antibiotic therapy, he was re-evaluated at an outpatient ID clinic with complete resolution of his mid-thoracic and rib pain with stabilization of his chronic low back pain. Repeated outpatient labs obtained demonstrated a WBC of 5.35/mm3 and an improved CRP of 8.8 mg/L. He was discharged from the ID clinic given both clinical and serological improvement and finished his six-week course of IV vancomycin.

Discussion

Based on literature review, the connection between *S. caprae* and BJIs in humans was first reported in 1997, and 55 cases of *S. caprae* BJIs have been published worldwide [1,2,4,6–8]. Two major studies have investigated the incidence of *S. caprae* BJIs [1,6–8].

The first study by d'Ersu et al. was a retrospective study reviewing bone and joint samples between January 2004 and March 2012 at Nantes University Hospital; they identified 13 patients with *S. caprae* BJIs and only four had infections involving their native bone – two with diabetic foot infections, one with recurrent osteomyelitis, and one with chronic osteitis [1,7]. In this study, a few *S. caprae* isolates were misidentified by traditional microbiology techniques and MALDI-TOF MS was found to be far superior in identification [1].

The second study by Seng et al. was a retrospective study that analyzed isolates from 2006 to 2012 at the University of Marseille and Nimes and found 96 % of cases were localized to the lower limbs – nine cases in the knee (36 %), four cases in the hip (16 %), one case in the femur (4 %), four cases in the tibia (16 %), three cases in the ankle (12 %), three cases in the foot (12 %) [8]. In this study, 88 % of *S. caprae* BJIs involved orthopedic device related infections; however, only 12 % occurred in the absence of orthopedic hardware. All 25 *S. caprae* BJIs strains were susceptible to vancomycin, teicoplanin, and doxycycline while 96 % of cases were also susceptible to methicillin, clindamycin, moxifloxacin, gentamicin, tobramycin, and co-trimoxazole [8].

Since the larger analyses by Seng et al. and d'Ersu et al. in 2014 and 2016, respectively, three additional *S. caprae* publications involve BJIs have been released. Hilliard et al. in 2017 describes a 69-year-old male with a past medical history significant for cryptogenic (decompensated) liver cirrhosis, diabetes mellitus, a prior lumbar spine hemi-laminotomy, and a recently completed course of antibiotics (initially ertapenem, then vancomycin) for L4-L5 discitis and bacteremia with *S. caprae* (MIC $<$ 1 mg/dL) three months prior who was seen at another emergency department prior for acute on chronic back pain and lower extremity weakness and pain [7]. Upon admission initial lab findings noted a WBC of 9200/mm^3, ESR 83 mm/hr, and CRP 2.5 mg/dL. MRI imaging noted L4-L5 spinal discitis/osteomyelitis with epidural phlegmon. Given clinical patient's clinical stability at the time, empiric antibiotics were deferred while biopsy and cultures were obtained. Unfortunately, the patient rapidly decompensated with hepatic failure, severe coagulopathy, acute kidney injury, and gross hemoptysis. Blood cultures from the ED visit prior to admission noted *S. caprae* with MIC of 4 mg/dL (intermediate resistance). ID was consulted and recommended the addition of piperacillin/tazobactam, vancomycin, and doxycycline to his current cefazolin regimen. Significant amount of supportive of care was administered; however, patient ultimately expired.

In 2020, Fan et al. report a case of a 65-year-old male with acute on chronic back pain radiating to his left lower extremity [5]. Interestingly, the patient received acupuncture treatment, with the longest needle measuring approximately 7–8 cm of his lower back multiple times in the month prior to admission. Reportedly, the patient subjectively felt the needle contact his bone. Additional CT imaging noted vertebral damage, sequestrum formation, and hyperplasia of L4-L5 vertebrae; MRI signaled inflammation at L4-L5 level as well. Fluoroscopy guided needle puncture confirmed *S. caprae* growth [5]. Patient recovered well long-term after four weeks of vancomycin then two weeks of linezolid. Lastly, Vazquez et al. in 2023 highlight a case of subacute *S. caprae* osteomyelitis in a toe phalanx of a teenage girl [9]. Initial lab findings were not highly abnormal with a WBC 7600/mm^3, CRP less than 0.3 mg/L, and

ESR 7 mm/hr. No specific inciting event or risk factors were discussed and noted. The patient recovered well after a short course of oral antibiotics and subsequent debridement and curettage of the infected area [9].

Our case highlights a rare presentation of an emerging commensal pathogen (*Staphylococcus caprae*) as the cause of native thoracic osteomyelitis in an otherwise immunocompetent patient. This organism is closely related to *S. epidermidis* and *S. capitis* at the species level based on comparative genomic analysis and can form persistent biofilms leading to invasive BJIs in humans [1,3,8,10]. *S. caprae* vertebral discitis/osteomyelitis is clinically and radiologically indistinguishable from other spinal infections caused by *Staphylococcus aureus* or CoNS species. Due to advances in molecular identification, *S. caprae* can be better recognized through a combination of biochemical tests including strongly positive DNase and pyrrolidonyl aminopeptidase reactions and acid production from maltose [4,10].

Our patient had a history of diet-controlled diabetes mellitus and end-stage renal disease (ESRD) on hemodialysis (HD) with a prior catheter-related bloodstream infection (CRBSI) secondary to CoNS pathogen (*S. caprae*). Based on the 2009 IDSA guidelines for uncomplicated CRBSIs, he was treated appropriately with systemic antibiotics for one week given that his vascular catheter was removed with no active malignancy or immunosuppression [11]. His new dialysis access served as a potential source and biofilm development, leading to a recurrent CRBSI and subsequent hematogenous seeding into his vertebral spine. This could explain the development of an invasive CoNS spinal infection in an otherwise immunocompetent adult. His thoracic bone biopsy confirmed *S. caprae* osteomyelitis. Treatment for CoNS BJI consists of vancomycin 15 mg/kg IV every 12 hours to achieve trough level of 15–20 mcg/mL (or cefazolin for methicillin-susceptible *Staphylococci*) with consideration of rifampin 300 mg IV/PO every 8 hours for hardware-associated infections to target biofilm production; duration of therapy for CoNS BJI (or complicated CRBSI involving osteomyelitis) is 6–8 weeks in adults [11]. Our patient had his dialysis catheter replaced after clearance of his *S. caprae* bacteremia and completed a six-week course of intravenous vancomycin with outpatient HD sessions.

Interestingly these recent *S. caprae* cases, including ours, did not present with overt septic symptoms or a high degree of acute symptoms which makes the detection of *S. caprae* challenging. Many of which also had relatively normal inflammatory markers as well. Notably our patient did not receive trauma (i.e. acupuncture) or was immunocompromised (i.e. decompensated cirrhotic) compared to Fan et al. or Hilliard et al.; however, our patient did have a direct portal of entry with a dialysis catheter. *S. caprae* should be recognized as an emerging virulent pathogen in immunocompetent patients despite the absence of orthopedic prostheses. This organism can lead to both community acquired and nosocomial invasive spinal infections and prompt treatment can prevent residual neurological deficits and unfavorable outcomes.

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Author statement

All authors have reviewed and approved the new revised manuscript for re-submission.

Conflict of interest

The contents of this article do not represent the views of the Department of Veterans Affairs or the United States Government. The interpretation and reporting of this case are the responsibility of the authors and in no way should be seen as official policy or interpretation of the United States Government.

The authors whose names are listed on this publication certify that they have no affiliations or involvement in any organization or entity with any financial interest or non-financial interest in the subject matter or materials discussed in this manuscript. They also certify all information presented is their own opinion and not of their employers.

Consent

A consent was obtained from the patient for publication of this case report and accompanying images.

Ethical approval

N/A.

CRediT authorship contribution statement

Peter J Skidmore: Writing – review & editing, Supervision. **Stephanie L Baer:** Writing – review & editing, Supervision. **Patrick J Tu:** Writing – review & editing. **Zoheb Irshad Sulaiman:** Writing – review & editing, Writing – original draft, Visualization, Conceptualization.

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.idcr.2024.e01962.

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