Introduction.

Staphylococcus caprae is a coagulase-negative staphylococcus that has been reported in several cases as a human pathogen. However, it has rarely been reported as pathogen in native bone. Furthermore, the reported MIC levels noted in the literature for vancomycin were <2 µg ml⁻¹ making vancomycin a first line choice for infected patients.

Case presentation.

We report a case of Staphylococcus caprae causing osteomyelitis of the lumbar spine and bacteraemia and resulting in sepsis and ultimately the demise of a patient despite appropriate prolonged antibiotic therapy.

Conclusion.

Staphylococcus caprae has been reported as a human pathogen since 1983 when it was discovered. We report a case involving native bone infection which is rare in the absence of mechanical hardware. Furthermore, this strain had an elevated MIC for vancomycin which has not been reported in the literature.
examination, the patient had no fevers and had normal vital signs. He did not have point tenderness on his back, and had 2/5 strength documented in all his extremities, normal sensation and no documentation of gait. Laboratory tests revealed a normal white blood cell count of 9.2 K per mm$^3$, sedimentation rate of 83 mm h$^{-1}$, and C-reactive protein of 2.5 mg dl$^{-1}$, with evidence of an acute kidney injury (serum creatinine level: 2.1 mg dl$^{-1}$ with a normal baseline) and decompensated liver cirrhosis. Magnetic resonance imaging (MRI) of the lumbar spine revealed spinal discitis/osteomyelitis at L4–L5 level (Fig. 1). Initially, antibiotics were held as the patient was still haemodynamically stable and a biopsy of affected spine lesion with cultures was desired to establish a microbiological diagnosis.

The patient rapidly developed progressive hepatic failure with severe coagulopathy and acute kidney injury after hospital admission. On hospital day 4, he developed gross haemoptysis with increasing oxygen requirements and was transferred to the medical intensive care unit. Chest x-ray revealed diffuse patchy opacities bilaterally, that were not present on admission. The patient was intubated and a bronchoscopy was performed showing evidence of diffuse alveolar haemorrhage. Blood cultures drawn at the outside hospital emergency department immediately prior to admission were growing *Staphylococcus caprae* with intermediate resistance to vancomycin documented in the report sent to our hospital (established by Vitek 2 at outside hospital, confirmed by manual MIC testing, although MIC result was 4 µg ml$^{-1}$ per verbal report). The patient went into shock and was started on vasopressors. He was started on cefazolin for coverage of *Staphylococcus caprae* at the recommendation of our Infectious Disease specialists, in addition to vancomycin, piperacillin-tazobactam and doxycycline for additional coverage for possible infectious aetiologies for diffuse alveolar haemorrhage. Despite aggressive antimicrobial therapy and supportive care, the patient’s haemodynamic status continued to deteriorate and the patient expired on hospital day nine.

Autopsy revealed necrotic and haemorrhagic L4–L5 vertebral bodies and disc spaces, with evidence of bilateral pleural effusions with diffuse alveolar haemorrhage. Tissue cultures from the affected vertebral bodies grew *Staphylococcus caprae* with MIC of 4 µg ml$^{-1}$ for vancomycin established with automated microdilution method (Vitek 2; bioMérieux) and confirmed with Etest (bioMérieux).

**DISCUSSION**

*Staphylococcus caprae* has been noted as a human pathogen since the late 1980s after its discovery in 1983. There are reports in the literature of a variety of infections caused by *S. caprae* but by far the largest number of infections occur in bone and joint, and of these, the vast majority are in patients with orthopaedic devices in place. *S. caprae* has been uncommonly reported as a pathogen of native bone and here we report a case of native bone infection as the source of bacteraemia. No prior studies have shown a MIC of 4 µg ml$^{-1}$ for vancomycin for *S. caprae*; prior studies publishing MICs are limited but were <2 µg ml$^{-1}$ and recommended vancomycin as first line therapy against *S. caprae* [10]. At our institution, doxycycline was a viable alternative and cefazolin was chosen due to the consistent susceptibility of this individual’s *S. caprae* isolates to oxacillin (cephalosporins are not routinely tested when organism is a species of the genus *Staphylococcus*). Table 1 lists the three separate cultures of *S. caprae* during this individual’s clinical course as well as their MIC values. Interestingly, vancomycin began as MIC of 1 µg ml$^{-1}$ and then further MIC values were 4 µg ml$^{-1}$.

Several case reports of infections related to *S. caprae* are available in the literature. Table 2 lists most of the available case reports. There was also a case report that mentioned a 72-year-old Japanese man with recurrent *S. caprae* sepsis but this report was only available in Japanese [5]. A different study out of Japan did identify *S. caprae* in the urine of patients who had received chemotherapy but it was not evident whether it was infectious or not. However, in that report they did note a high incidence of methicillin resistance (11% of all methicillin-resistant organisms were *S. caprae*), and that *S. caprae* totalled 6% of all coagulase-negative staphylococcal species isolated [8]. In 2004, Ross and colleagues published a study in which 10 of 36
neonates in their neonatal intensive care unit were colonized with *S. caprae*, of which six cases were bacteraemic, one case was a cerebrospinal fluid (CSF) shunt infection and one was a vascular catheter-associated infection, and it was noted that 13% of the *S. caprae* strains recorded carried the mecA gene [3].

There are two major studies looking at the incidence of *S. caprae* bone and joint infections. The first, by d’Ersu et al. [6], was a retrospective study done at Nantes University looking at data between 2004 and 2012. They identified 13 patients with *S. caprae* bone and joint infections, and in this study four patients had infection of their native bone: two individuals with diabetic foot infections, one with recurrent osteomyelitis and one with chronic osteitis [6].

The second study was published by Seng et al. [4] and looked at two major hospitals in France between 2006 and 2012, the University of Nantes and the University of Nimes. The University of Marseille had 16 bone and joint infections from 2006 to 2012 caused by *S. caprae* and the University of Nimes had nine bone and joint infections from 2007 to 2012 caused by *S. caprae*. In this study, only one patient had infection of a vertebra, and only three patients had no orthopaedic devices present at the time of infection. Most patients required more than two antibiotics with surgical debridement and removal of the orthopaedic device for complete clearance of infection [4].

Interestingly, a study published in 2014 looking at small colony variations in bone and joint infections found that in 76 human samples only two grew *S. caprae*/*Staphylococcus capitis* and that distinguishing between the two can be difficult when using the 16S rRNA gene, which is considered the gold standard [11, 12]. Furthermore, in the study of d’Ersu et al. from 2016 they found that a small number of isolates

| Outside hospital March 2016 (initial, blood): | Outside hospital July 2016 (blood): | Our institution July 2016 (lumbar spine): |
|---------------------------------------------|------------------------------------|----------------------------------------|
| Levofloxacin <0.5                           | Levofloxacin 0.5                    | Erythromycin 1                          |
| Clindamycin <0.25                          | Clindamycin 0.25                    | Oxacillin <0.25                         |
| Erythromycin <0.25                         | Erythromycin 1                      | Tetracycline <1                         |
| Oxacillin <0.25                            | Oxacillin <0.25                     | Bacitracin <10                          |
| Tetracycline <1                            | Tetracycline <1                     | Vancomycin 1 Int (verbal =4)           |
| Bacitracin <0.5                             | Bacitracin <0.5                     | Vancomycin 4                           |
| Vancomycin 1                               | Vancomycin 10                       | Rifampin <0.5                           |
| Penicillin >10 R                           | Erythromycin 1                      | Doxycycline <0.5                        |
| Ciprofloxacin <1                           | Erythromycin 1                      | Oxacillin <0.25                         |
| Meropenem <2                               | Erythromycin 1                      | Bacitracin <0.5                         |
| Gentamicin <1                              | Erythromycin 1                      | Vancomycin <10                          |

Table 2. Summary of case reports involving *S. caprae* infections

| Author and year of case(s) | Publication type | Site of infection | Clinical history |
|----------------------------|------------------|-------------------|------------------|
| Vandenesch *et al.* 1988–1992 [9] | Case series report | UTI | 46-year-old s/p medullary decompression for rhabdomyosarcoma |
| Shuttleworth *et al.* 1990–1996 [10] | Case series report | Bone and joint infection | 9 cases, 7 of which were traumatic fractures |
| Kanda *et al.* 1998–2000 [13] | Case series report | Bacterial orbit externa | 2.9% of 202 cases caused by *S. caprae* |
| Devrise *et al.* and Barelli *et al.* 1999 [2, 14] | Scientific review articles | Nosocomial infections, varied | 3 of 53 cases were caused by *S. caprae* |
| Benedetti *et al.* 2008 [5] | Case report | CSF infection | 47-year-old female with lumbar-sacral s/p spinal analgesia pump implantation, required device removal and 6 weeks IV antibiotics |
| Kato *et al.* 2010 [8] | Case report | Bacteremia | Neutropenic patient after induction chemotherapy for ac, associated with central line |
| Shin *et al.* 2011 [15] | Case report | Peritonitis | 3 patients receiving peritoneal dialysis |
| Henry *et al.* 2014 [7] | Case report | Endophthalmitis | 24-year-old healthy female after surgery for vitreous floaters |
were misidentified by a commercial identification system. MALDI-TOF MS was found to be far superior in identification rates of *S. caprae* [6].

For our individual patient, both failure of antibiotic therapy and source control likely contributed to his ultimate clinical outcome. In the available literature it is clear that without removal of infected bone or devices, antibiotic clearance of the infection did not occur. Unfortunately, given his comorbidities and rapid clinical decline, our patient was not a surgical candidate. Additionally, production of toxins by this strain of *S. caprae* was not tested.

In conclusion, *S. caprae* is still an understudied organism of human infection, and it has been determined to carry the *mecA* gene conferring methicillin resistance making it an important pathogen to be identified early on, especially in patients with significant comorbidities.

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**Conflicts of interest**

The authors declare that there are no conflicts of interest.

**Ethical statement**

The authors complied with institutional ethics policies in the writing of this case report.

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