A Rare Case of Staphylococcus caprae-Caused Periprosthetic Joint Infection Following Total Hip Arthroplasty: A Literature **Review and Antibiotic Treatment Algorithm Suggestion** Philip Domashenko ^{1,2}, Georgios Foukarakis ^{1,2}, Eustathios Kenanidis ^{1,2}, Eleftherios Tsiridis ^{1,2}

Background

A common pathogen in both community and hospital infections, Staphylococcus caprae (S. caprae) is a commensal, coagulasenegative Staphylococcus found in the skin flora of goats and humans [1]. S. caprae is capable of creating biofilms on prosthetic materials and adhering to human tissues via fibronectin-binding proteins, successfully evading antibiotics and the body's natural chemotactic leukocyte response [2,3].

S. caprae infections have a similar clinical presentation to other Staphylococcus pathogens. Symptoms such as fatigue, pain, swelling, redness, and warmth at the incision site are typical for periprosthetic joint infections. In the literature 413 S. caprae isolates have been documented globally since the first reported case in 1997 [1], including 55 cases of bone joint infections. It occurs predominantly in the lower limb joints. In summary, including the current case, 10 cases of PJI of the hip have been described in the literature, making them an extremely uncommon condition.

This study highlights a very rare case of PJI infection due to S. caprae, reviews the limited literature, and provides the available evidence for surgical and antibiotic management.

Case Presentation

A 59-year-old male patient underwent a primary uncemented THA in June 2022 due to right hip osteoarthritis (Figure 1). After two days, the patient was discharged without complications (Figure 1). In September 2022, the patient presented febrile (39°C) with groin and buttock pain. The physical examination revealed swelling, redness, and high temperature in the surgical incision area. The results of the laboratory tests showed an ESR of 45 mm and a C-reactive protein (CRP) level of 3.9 mg/dL. The MRI showed a pathologically increased liquid concentration around the right hip joint with bone edema (Figure 2).

Debridement, antibiotics, and implant retention (DAIR) was chosen as the most effective treatment since it was strongly suspected that the prosthetic joint was infected. Synovial fluid was aspirated in the operating room, and six biopsy samples were collected for cultures. All infected tissue was removed by meticulous debridement and irrigation with pulsed lavage, and the removable parts were replaced. Four out of six cultures came positive for S. caprae, which was sensitive to most antibiotics.

The patient was treated with 1 g intravenous (IV) vancomycin and 400 mg IV ciprofloxacin twice a day for 12 days as an inpatient, followed by 500 mg oral ciprofloxacin and 300 mg oral clindamycin three times a day as an outpatient for three months of antibiotic treatment.

Despite the remission of symptoms and reduced inflammatory markers to normal levels on discharge, the patient was readmitted in December 2022 febrile (38° C) with the same clinical presentation. At the time of admission, the level of CRP was 10.7 mg/dl and ESR 45mm. This time a full prosthesis revision was performed. During the operation, synovial and tissue samples were collected for cultures which were negative as expected due to prior antibiotic treatment. The femoral stem and acetabular component were removed, and thorough debridement and irrigation with pulse lavage were performed. Vancomycin-loaded bone cement (1 gr per 40 mg of cement) was used, and a fully cemented prosthesis was placed (Figure 2). The patient was hospitalized for 10 days and treated with 1 gr IV Vancomycin twice a day and 2 gr IV Cefoxitin three times a day until the inflammatory markers normalized. This was, followed by 11 days of outpatient 1 gr IV Vancomycin and 500 mg IV Levofloxacin twice a day administration. After completing three weeks of intravenous administration, he continued with 500 mg oral Levofloxacin and 600 mg oral Rifampin twice daily for three months. Forthcoming, we will proceed to the second-stage revision.

S. caprae is a rare pathogen that occurs predominantly in the lower limbs and often affects prosthetic joints, as in this case. There are few reported cases due to the lack of rapid and accurate diagnostic tools and, more likely, a lack of awareness in clinical laboratories and its absence from more commonly used commercially available databases [4,5]. Several diagnostic methods have been used to identify S. caprae, such as Vitek, ribotyping 16S-23S printer, soda gene sequencing, and ID32 Staph. Matrix-assisted laser desorption/ionization time-of-flight mass spectrometry (MALDI-TOF MS) is a fast and reliable method, with an identification rate of 97%, for identifying bacteria from agar media [6]. Direct identification from positive blood cultures should decrease the time to obtain the result. In our case, preoperative joint aspiration came up positive for S. caprae infection. Diagnosis confirmation was made by cultures examination of the obtained tissue samples.

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Discussion



Figure 1. Preoperative anteroposterior plain radiograph of the pelvis The image shows right hip osteoarthritis. Note joint space narrowing and articular surface sclerosis (left). Postoperative anteroposterior plain radiograph of the pelvis. The image shows an uncemented right THA with titanium femoral stem implant and

ceramic-on-polyethylene bearing surfaces (right).

There is no agreement on the appropriate surgical therapy strategy for patients with S. caprae PJIs due to the low incidence. Therapeutic protocols not specific for S. caprae are used, including irrigation and debridement with retention of the prosthesis [7], one-stage revision with an exchange of the entire prosthesis (femoral head, femoral stem, acetabular shell, and acetabular liner) [8], two-stage revision with an exchange of the entire prosthesis [9], major partial one-stage revision (removal of either the acetabular shell or femoral stem) [10], or minor partial onestage revision (where only the femoral head and/or acetabular liner are exchanged) [1]. Seng et al. performed four one-stage and five two-stage revisions out of nine patients [11]. Mencia et al. suggest a one-stage revision with satisfactory results [12]. In our case, we performed DAIR followed by a two-stage revision with a vancomycin-loaded cemented spacer as the first stage. Four months down the line, the patient is free of symptoms, and we will proceed to the second stage.

Study	Treatment combinations	Antibiotics	Use (%)	Area of infection
Seng et al. [11]	Fluoroquinolone + rifampin	-	18.1	-
Achermann et al. [15] Darrieutort- Laffite et al. [16]	Fluoroquinolone + rifampin Fluoroquinolone + rifampin	Ciprofloxacin + rifampin Ofloxacin + rifampin	25.4 1.8	Elbow PJI Sacroiliac joint
Mencia et al. [12], Achermann et al. [5], Pommepuy et al. [17]	Quinolone + rifampin	Levofloxacin + rifampin	5.4 (hip), 7.2 (elbow	Hip PJI, elbow PJ
Achermann et al. [15]	Fluoroquinolone	Ciprofloxacin	1.8	Elbow PJI
Seng et al. [11]	Glycopeptide + fluoroquinolone + rifampin	Vancomycin + rifampin + fluoroquinolone	10.9	-
Seng et al. [11]	Carbapenem + glycopeptide	Vancomycin + imipenem	1.8	2
Seng et al. [11]	Glycopeptide + rifampin + fusidic acid	Teicoplanin + rifampin + fusidic acid	1.8	~
Seng et al. [11]	Glycopeptide	Vancomycin	3.6	-
Achermann et al. [15]	Penicillin + beta- lactamase + rifampin	Amoxicillin- clavulanate + rifampin	1.8	Elbow PJI
Darrieutort- Laffite et al. [16]	Penicillin + rifampin	Oxacillin + rifampin	1.8	Knee PJI
Achermann et al. [15], Elsner et al. [18]	Penicillin	Amoxicillin	3.6 (elbow), 1.8 (knee)	Elbow PJI, knee after an ACL repair
Seng et al. [11]	Sulfonamides + fluoroquinolone + rifampin	Co-trimoxazole + fluoroquinolone + rifampin	1.8	
Seng et al. [11]	Sulfonamide + fluoroquinolone	Co-trimoxazole + fluoroquinolon	1.8	-
Seng et al. [11]	Sulfonamide + tetracycline	Co-trimoxazole + doxycycline	1.8	*
Seng et al. [11] Achermann et al. [15]	Sulfonamides Fusidic acid + rifampin	Co-trimoxazole Fucidin + rifampin	1.8 1.8	Elbow PJI
Seng et al. [11]	Tetracycline	Doxycycline	1.8	
Achermann et al. [15]	Oxazolidinones	Linezolid	1.8	Elbow PJI

Table 1: Antibiotic treatment combinations use (%). ACL: anterior cruciate ligament; PJI: periprosthetic joint infection

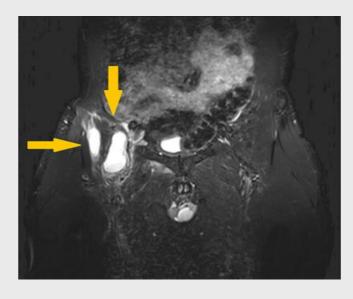




Figure 2.T1-weighted magnetic resonance imaging coronal plane of the pelvis. The image shows a pathologically increased liquid concentration and bone edema around the right hip joint (left).

Anteroposterior plain radiograph of the pelvis. The image shows a temporary antibiotic-loaded cement spacer (right).

With an emphasis on patient-reported outcomes for pain and function as well as reinfection rates and cost efficacy, several studies have compared one-stage and two-stage revision [13]. The gold standard was a two-stage revision since it had the lowest incidence of reinfection [14].

Antibiotic treatment in our case included oral levofloxacin and rifampin after IV vancomycin and levofloxacin. Table 1 describes the antibiotic treatment strategies of six articles and 55 cases of S. caprae PJIs, including seven hip joints. Overall, 35 cases received two antibiotics, nine received three antibiotics, and nine received one antibiotic. In most cases (43.5%), fluoroquinolone + rifampicin was used, followed by glycopeptide + fluoroquinolone + Rifampicin (10.9%). We suggest a double antibiotic treatment which consists of one fluoroquinolone/glycopeptide/quinolone/penicillin or β-lactam combined with rifampin.

Conclusions

S. caprae is a coagulase-negative Staphylococcus that is extremely uncommon and has a low frequency in the general population. Despite this, it is still an emerging human pathogen in patients with orthopedic infections. There have been only 10 hip PJIs described globally, making them very rare. In the future, identification tools like MALDI-TOF MS, which have demonstrated promising results, will add new data. The therapeutic alternatives are taken from non-S. capraespecific PJI protocols. For a successful clinical outcome, we advise early detection and removal of all implants to control infections embedded in biofilm. Two-stage revision is the gold standard surgical treatment for this type of PJI. For antibiotic treatment, one fluoroquinolone/glycopeptide/quinolone/penicillin or β-lactam should be used in conjunction with rifampicin for better results.

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