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Case report

# A rare case of *Staphylococcus caprae*–infected total hip arthroplasty: a report and literature review

Marlon Mencia [a], Shamir O. Cawich [a, *], Jacinta Bronte-Tinkew [b]

[a] Department of Clinical Surgical Sciences, University of the West Indies, St. Augustine Campus, West Indies, Trinidad and Tobago
[b] Center for Scientific Review, National Institutes of Health, 6701 Rockledge, Drive, Room 3164, MSC 7770, Bethesda, MD 20892, USA

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ABSTRACT

We report a rare case of *Staphylococcus caprae* hip arthroplasty infection. To be best of our knowledge, this is the 38th case of *S. caprae* prosthetic joint infection and the 9th case of *S. caprae* hip arthroplasty infection to be reported in the medical literature. The case was successfully treated by minor partial one-stage revision and culture-directed antibiotic therapy. Clinicians must maintain a low index of suspicion for *S. caprae* as a pathogen in orthopedic infections. We suggest that a minor partial one-stage revision is appropriate treatment because it balances low morbidity with good functional outcomes and a low chance of re-revision for infection at two years. We propose the following as markers of therapeutic success: (1) early detection of infection, (2) absence of radiolucent lines around the bone–prosthesis interface on plain radiographs, (3) monomicrobial infection, (4) infection with an organism of low virulence, (5) culture-proven susceptibility to available antibiotics, and (6) immunosuppression that is effectively treatable.



## 1. Background

Total hip arthroplasty (THA) improves quality of life and restores function to many patients with degenerative hip disease. Approximately 1% of these patients will develop prosthetic joint infections (PJIs) after the primary procedure.[1] This may cause implant failure and usually requires revision arthroplasty.[1,2] It may lead to multiple operations, long periods of patient disability, suboptimal outcomes, and increased mortality.[3]

The commonest microbes to be isolated in patients with PJIs are *Staphylococcus aureus* and *Staphylococcus epidermidis*,[4] but *Staphylococcus caprae* is a rare cause of orthopedic infections.[5] We now report the 9th recorded case in which *S. caprae* was isolated and the cause of an infected THA.

## 2. Report of a case

A 50-year-old man with no comorbidities presented with disabling right hip pain and underwent an uncomplicated THA. At postoperative week 8, he presented with low-grade pyrexia and pus draining from the skin incision (Fig. 1). He denied experiencing pain but had a pronounced Trendelenburg gait. Blood tests revealed elevated levels of C-reactive protein (CRP) at 7.1 mg/dl (normal range, 0.010–1.0 mg/dl) and erythrocyte sedimentation rate (ESR) at 104 mm (normal range, 0–20 mm). On plain radiographs (Fig. 2), there was subcutaneous emphysema noted around the surgical scar, but it did not appear to track deep down to the prosthesis or periprosthetic tissue planes.

The patient was resuscitated, and empiric intravenous vancomycin was commenced after wound swabs and blood samples were taken for cultures. With a suspicion of PJI, he was prepared for anesthesia and taken to the operating room. With the patient in the left lateral position, a modified direct lateral (Hardinge) approach was used with a 10-cm incision made behind the greater trochanter and in line with the femur. After evacuation of pus, the abscess cavity was found to extend deep into the fascia. The abductors at the greater trochanter had dehisced, and pus tracked down toward the prosthesis. Biopsy samples were taken for tissue culture from six sites around the prosthesis. This was followed by aggressive debridement and irrigation of the periprosthetic tissues (Figs. 3–4). The modular components (femoral head and acetabular liner) were exchanged at the same sitting by minor partial one-stage revision. The abductors were repaired using 2/0 polyglactin

* Corresponding author. Fax: +1-876-620-4560.
*E-mail address:* Shamir.Cawich@sta.uwi.edu (S.O. Cawich).

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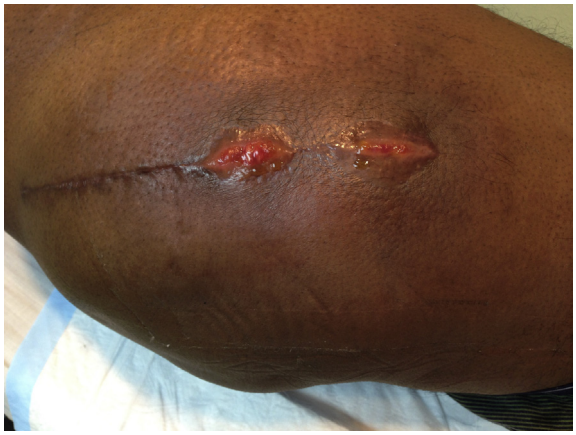

**Fig. 1.** Photograph of the surgical site infection at postoperative week 8. The wound has dehisced, and pus has been evacuated.

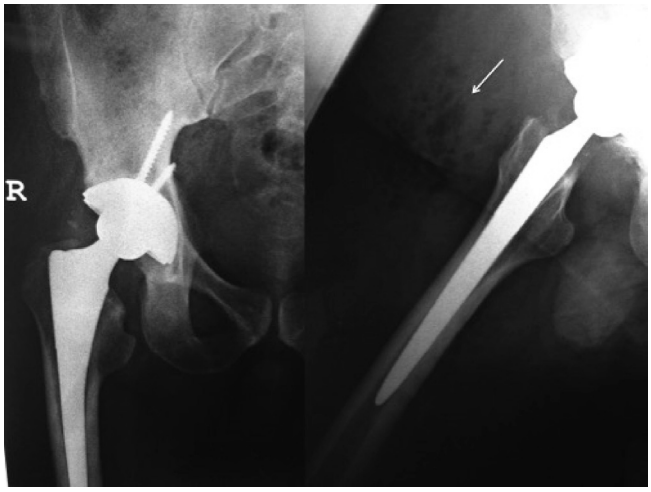


**Fig. 2.** Plain radiographs of the replaced hip. Subcutaneous emphysema is present (arrow), but there are no radiolucent lines around the femoral stem or acetabular shell.

sutures using intraosseous tunnels, and the operative wound was closed in layers.

The deep tissue samples were cultured on sheep blood and MacConkey agar plates and incubated at 37 °C under aerobic and anaerobic conditions for 10 days. Gram staining and catalase/oxidase testing were used to identify the organisms. Our laboratory confirmed the presence of *S. caprae* using the Vitek 2 system (BioMerieux) four days later. All six tissue cultures were positive for *S. caprae*, which was sensitive to levofloxacin and rifampicin. At this point, we commenced 750 mg of intravenous levofloxacin and 300 mg of intravenous rifampicin twice a day. Regular liver function and renal function tests were performed to exclude organ injury.

During his hospitalization, there were multiple elevated serum glucose readings and an elevated $HB_{A1C}$ level at 9%, although there was no prior history of diabetes mellitus. He was considered to have a new diagnosis of diabetes mellitus, requiring insulin and oral hypoglycemics for appropriate glycemic control.

There were no complications noted during the recovery period. He was discharged from the hospital on day 14 after operation after being converted to the equivalent oral dose formulation of levofloxacin and rifampicin. Over the course of three months, the antibiotic doses were reduced to 500 mg of levofloxacin and 150 mg of rifampicin twice daily, in accordance with decreasing CRP and ESR levels. During this time, liver function and renal function

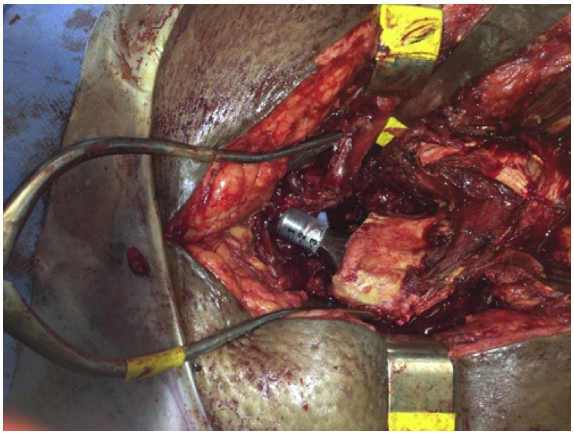

**Fig. 3.** Intraoperative photograph after evacuation of the pus. The femoral head has been removed, and the neck is now covered by a 3-cc syringe to protect the morse taper.

tests were performed weekly to ensure that there was no organ damage.

During outpatient surveillance, there was a consistent downward trend in CRP and ESR levels, when measured on a two-weekly basis. In addition, there was better glycemic control, with a reduction in the $HB_{A1C}$ level to 5.7%. Postexploration radiographs showed no evidence of loosening (Figs. 1 and 2).

After three months of outpatient antibiotic therapy, there was normalization of CRP levels to 0.8 mg/dl, ESR to 18 mm/hr, and glycemic control with $HB_{A1C}$ at 5.2%. At this point, the patient ambulated unaided and without pain. He did have a mild Trendelenburg gait but was active and able to return to work. To date, there have been no signs of recurrent infection up to 30 months after revision.

## 3. Discussion

*S. caprae* is a gram-positive bacillus belonging to the *Staphylococcus spp*. It is a known commensal in goats[6] and sheep[7] but it is an uncommon pathogen in human infections.[5,7,8] In cases where it is isolated as a human pathogen, *S. caprae* has been reported to cause urinary tract infections,[9] endocarditis,[10] meningitis,[11] otitis externa,[9] bacteremia,[9] epidural catheter infections,[11] chemotherapy reservoir infections,[12] and dialysis catheter infections.[13]

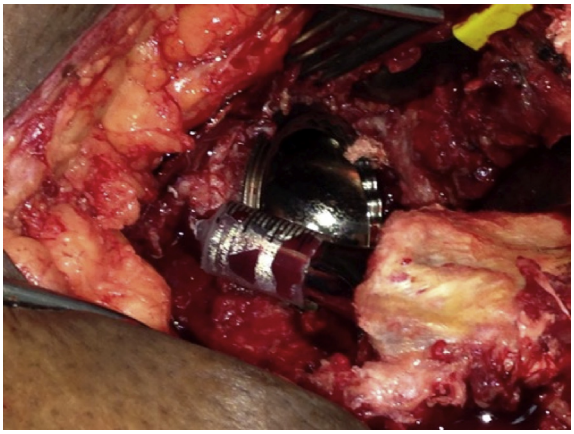

**Fig. 4.** The modular components (acetabular liner and femoral head) have been explanted. The femoral stem and acetabular shell remain in situ. This will facilitate proper debridement and irrigation before the minor partial one-stage replacement.

There have also been reports of orthopedic infections at prosthetic joints,[5,14] although *S. caprae* still remains a rare cause of orthopedic infections. Arciola et al.[15] examined 1311 isolates of staphylococcal species isolated from patients with orthopedic infections. The majority of isolates were *S. aureus* (47%) and *S. epidermidis* (36%). *S. caprae* only accounted for 0.4% of the staphylococcal species causing orthopedic infections.[15]

There is a predilection for *S. caprae* PJIs to occur in males at a mean age of 58 ± 17 years[14] in those with the body mass index >30, concomitant malignancies,[14] diabetes mellitus,[14] and taking immunosuppressive therapy/corticosteroids.[14] Our patient fits this profile well, and he also had a new diagnosis of diabetes mellitus made during the postoperative recovery period.

Although it is a recognized animal commensal, only 20% of humans who develop *S. caprae* infections have any history of animal contact.[13] Similarly, in our case, there was no history of recent animal contact or high-risk activity. We believed this to be a hospital-acquired infection. This is in keeping with the prevailing theories in the medical literature that *S. caprae* infections are mostly nosocomial infections.[9,16]

Most *S. caprae* orthopedic infections occur in patients with indwelling prosthetic implants (60%).[14] Only 28% occur in patients with osteosynthesis devices, and 12% occur in patients without orthopedic devices.[14] Even so, *S. caprae* PJIs are rare. Seng et al.[14] performed a review of the medical literature and reported that there were only 36 reported cases in the existing medical literature of confirmed *S. caprae* PJI up to December 2012. We performed a literature search in PubMed, Embase, Medline, and Google Scholar using the key words staphylococcus, caprae, prosthesis, joint, infection, and implant. We found only one additional report of prosthetic THA infection with *S. caprae* originating in France.[17] To the best of our knowledge, this is now the 38th case of *S. caprae* PJI to be reported in the medical literature.

When the search was narrowed to identify the infected THAs, the number of reported cases was even smaller. Of the 37 existing cases of *S. caprae* PJIs reported in the medical literature, only 8 were *S. caprae*–infected hip prostheses,[14,17–20] including the most recent reported by Pommepuy et al.[17] in France. In 16 cases, the prosthetic joint was not specified because these reports focused on the microbial properties of *S. caprae*. Despite attempts to contact the corresponding authors, these details remained unavailable. Therefore, to the best of our knowledge, this is now the 9th case of *S. caprae* THA infection reported in the medical literature.

This distinction is important because the pathophysiology of infection differs from that for osteosynthetic devices. The *S. caprae* osteosynthesis device infections tend to be polymicrobial,[14] and this is believed to be due to contamination from traumatic/dirty wounds at the time of implantation. However, 60% of PJIs tend to be monomicrobial, with *S. caprae* being the only organism to be isolated.[14] This was the case in our patient. In these cases, the prevailing theory is that these are nosocomial infections due to contamination at the time of implantation.

Some authorities have suggested that the low incidence of *S. caprae* PJI (38 cases in the past decade) may be due to inadequate methods used for laboratory detection.[21] It has been suggested that detection is improved when molecular techniques are used, such as modern matrix-assisted laser desorption ionization time-of-flight (MALDI-TOF) mass spectrometry.[14] MALDI-TOF mass spectrometry technique was reported to have higher than 98% accuracy to identify *S. caprae* strains.[14] Unfortunately, MALDI-TOF mass spectrometry was not available in our laboratory, but we used the Vitek 2 system, which has been reported to have 75% overall accuracy to identify *S. caprae*.[14] Equally important is the technique for identification; it is important for clinicians to take appropriate samples. Kamme and Lindberg[22] recommended taking deep-tissue biopsy samples from at least 5 areas around the prosthesis. Growth in <2 biopsy samples suggested contamination, while infection was only diagnosed when all samples yielded positive growth.[22] We adhered to these recommendations by taking 6 deep-tissue biopsy specimens at operation. Because all 6 samples taken for cultures yielded the same organism, it is unlikely that there was contamination as one would expect multiple different organisms to be present at different sites if contamination was present.

Because the incidence is very low, there is no consensus on the best therapeutic approach for patients with *S. caprae* PJIs. Therapeutic options are borrowed from protocols for PJIs that are nonspecific to *S. caprae*. Even with these, there is still controversy about the best of these therapeutic options:[1] irrigation and debridement with retention of the prosthesis,[2] two-stage revision with exchange of the entire prosthesis,[3] one-stage revision with exchange of the entire prosthesis (femoral stem, femoral head, acetabular liner, and acetabular shell),[4] major partial one-stage revision (removal of either the femoral stem or acetabular shell), or[5] minor partial one-stage revisions (where only the femoral head and/or acetabular liner are exchanged).

Irrigation and debridement is sometimes used when prostheses are retained in patients being treated for PJIs, but the success rates are low, ranging from 16% to 46%. Romano et al.[23] performed a systematic review of 710 patients who had debridement, irrigation, and prosthesis retention for PJIs across 14 publications over a period of 41 years. This management strategy yielded success rates of 46% when treating infected hip prostheses and 52% for infected knee prostheses at a mean follow-up of 53 months. Based on these outcomes, irrigation and debridement has a limited role.[23]

Whenever possible, a revision is the usual modality of treatment. Two-stage revisions were considered the gold standard because they brought the lowest rates of reinfection.[18,24] However, the low reinfection rates must be balanced against the resulting severe functional impairment.[18] Therefore, two-stage revisions are usually used for severe chronic infections. The thrust has been toward one-stage revisions because their functional outcomes are better and their reinfection rates are comparable.[18,24]

Leonard et al.[25] carried out a systematic review of 9 studies that compared one-stage vs two-stage revisions in patients with infected THAs. There was a trend toward better functional outcomes with one-stage revisions, but a statistically significant difference could not be demonstrated. Leonard et al.,[25] however, lamented about the poor quality of the data and the lack of randomized controlled trials. We eagerly await results of the INFORM trial,[3] a prospective randomized two-arm superiority trial currently underway that is designed to compare one-stage and two-stage revisions with a focus on patient-reported outcomes for pain and function as well as reinfection rates and cost efficacy. The INFORM trial[3] began enrolling participants in 2015 and is anticipated to be completed in 2017.

Engesaeter et al.[24] examined revision options in 906 patients with infected primary THAs over a period of 22 years. When revision options were compared against a 2-stage revision (which had the lowest reinfection rate as expected) and using the absence of re-revision for recurrent infection as the end point, they reported that a minor partial one-stage revision (exchange of the head and/or liner with retention of the fixed implant) yielded 76% success. Compared with the outcomes with those of a two-stage revision, a minor partial revision yielded a statistically lower relative risk of re-revision for any reason (risk ratio [RR] = 1.5 vs 4.1, P=<0.001) and re-revision for infection (RR = 4.1 vs 6.0, $P < 0.001$) than a major partial revision. A one-stage minor partial revision was recommended as the more robust procedure, and this was our procedure of choice.

We exploited some of the known pathophysiologic mechanisms of *S. caprae* in our case. It is known that *S. caprae* has the ability to adhere to human tissues by producing fibronectin-binding proteins[26] and induces biofilm formation on prosthetic materials,[27] effectively isolating itself from antibiotics and the native chemotactic leukocyte response. In our case, we extrapolated that the prosthesis–bone interface had not yet been violated because plain radiographs did not reveal any radiolucency around the prosthesis on his presentation. On this basis, we thought it unnecessary to remove the femoral stem and acetabular shell. But with the knowledge that *S. caprae* produced adhesion proteins, we exchanged the modular components (femoral head and acetabular liner) in our procedure. We also ensured that aggressive surgical debridement was performed to excise as much inflammatory tissue, scar, and biofilm as possible.

In this case, intravenous levofloxacin was used as a culture-directed antibiotic. We also added rifampicin as an adjunct based on the experience/recommendations in the available medical literature.[14] Rifampicin inhibits bacterial DNA-dependent RNA polymerase and had the advantage of good oral bioavailability, allowing us to continue adequate long-term outpatient therapy. Long-term culture-directed antibiotic therapy is necessary to avoid the development of resistance.[28] The existing medical literature supports use of the combination of a fluoroquinolone with rifampicin.[14,28]

We acknowledge that there is no consensus on the best therapeutic option for *S. caprae* PJI, but with no recurrent infection, low morbidity, and good functional outcomes after two years, we propose that a minor partial one-stage revision should be considered. Of course, we suggest that it should be limited to select cases where some of the following markers of therapeutic success can be identified: (1) early detection of infection, (2) absence of radiolucent lines around the bone–prosthesis interface on plain radiographs, (3) monomicrobial infection, (4) infection with an organism of low virulence, (5) culture-proven susceptibility to available antibiotics, and (6) the absence of immunosuppression that is not effectively treatable. Our patient was recently diagnosed with diabetes mellitus, but this could be controlled with medical management.

## 4. Conclusion

Although it is still rare, *S. caprae* is emerging as a human pathogen in patients with orthopedic infections. This case adds to the available literature as the 38th reported case of a PJI and the 9th case of THA infection with *S. caprae*. Therefore, clinicians must maintain a low index of suspicion for *S. caprae* as pathogens in orthopedic infections. Laboratory staff should be alerted to ensure adequate mechanisms are used to identify the presence of *S. caprae* in suspected cases.

We propose that a minor partial one-stage revision is effective treatment for *S. caprae* PJIs because it balances low morbidity with good functional outcomes and a low chance of re-revision for infection at two years. We suggest using the following criteria as markers of therapeutic success: (1) early detection of infection, (2) absence of radiolucent lines around the bone–prosthesis interface on plain radiographs, (3) monomicrobial infection, (4) infection with an organism of low virulence, (5) culture-proven susceptibility to available antibiotics, and (6) the absence of immunosuppression that is not effectively treatable.

## Conflicts of interest

The authors declare no conflict of interest.

## Author contributions

M.M., J.B.-T., and S.O.C. conceptualized the study and collected data, wrote the paper, reviewed the article for scientific accuracy, and authorized the final version of the article.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.cmrp.2019.09.008.

## References


1. Pulido L, Ghanem E, Joshi A, Purtill JJ, Parvizi J. Peri-prosthetic joint infection: the incidence, timing, and predisposing factors. *Clin Orthop Relat Res*. 2008;466(7):1710–1715.
2. Clohisy JC, Calvert G, Tull F, McDonald D, Maloney WJ. Reasons for revision hip surgery: a retrospective review. *Clin Orthop Relat Res*. 2004;429:188–192.
3. Strange S, Whitehouse MR, Beswick AD, et al. One-stage or two-stage revision surgery for prosthetic hip joint infection - the INFORM trial: a study protocol for a randomised controlled trial. *Trials*. 2016;17(90):2–7.
4. Toms AD, Davidson D, Masri BA, Duncan CP. The management of peri-prosthetic infection in total joint arthroplasty. *Br J Bone Joint Surg*. 2006;88(2):149–155.
5. Shuttleworth R, Behme RJ, McNabb A, Colby WD. Human isolates of Staphylococcus caprae: association with bone and joint infections. *J Clin Microbiol*. 1997;35(10):2537–2541.
6. Moroni P, Pisoni G, Antonini M, et al. Subclinical mastitis and antimicrobial susceptibility of Staphylococcus caprae and Staphylococcus epidermidis isolated from two Italian goat herds. *J Dairy Sci*. 2005;88(5):1694–1704.
7. Devriese LA, Poutrel B, Kilpper-Balz R, Schleifer KH. Staphylococcus gallinarum and Staphylococcus caprae, two new species from animals. *Int J Syst Bacteriol*. 1983;33:480–486.
8. Vandenesch F, Eykyn SJ, Bes M, Meugnier H, Fleurette J, Etienne J. Identification and ribotypes of Staphylococcus caprae isolates isolated as human pathogens and from goat milk. *J Clin Microbiol*. 1995;33(4):888–892.
9. Ross TL, Fuss EP, Harrington SM, Cai M, Perl TM, Merz WG. Methicillin-resistant Staphylococcus caprae in a neonatal ICU. *J Clin Microbiol*. 2005;43(1):363–367.
10. Spellerberg B, Steidel K, Lütticken R, Haase G. Isolation of Staphylococcus caprae from blood cultures of a neonate with congenital heart disease. *Eur J Clin Microbiol Infect Dis*. 1998;17(1):61–62.
11. Benedetti P, Pellizzer G, Furlan F, Nicolin R, Rassu M, Sefton A. Staphylococcus caprae meningitis following intraspinal device infection. *J Med Microbiol*. 2008;57(7):904–906.
12. Kato J, Mori T, Sugita K, et al. Central line-associated bacteremia caused by drug-resistant Staphylococcus caprae after chemotherapy for acute myelogenous leukemia. *Int J Hematol*. 2010;91(5):912–913.
13. Shin JH, Kim SH, Jeong HS, et al. Identification of coagulase-negative staphylococci isolated from continuous ambulatory peritoneal dialysis fluid using 16S ribosomal RNA, tuf, and SodA gene sequencing. *Perit Dial Int*. 2011;31(3):340–346.
14. Seng P, Barbe M, Pinelli PO, et al. Staphylococcus caprae bone and joint infections: a re-emerging infection? *Clin Microbiol Infect*. 2014;20(12):1052–1058.
15. Arciola CRR, Campoccia D, An YH, et al. Prevalence and antibiotic resistance of 15 minor staphylococcal species colonizing orthopedic implants. *Int J Artif Organs*. 2006;29:395–401.
16. Couto I, Pereira S, Miragaia M, Sanches IS, de Lencastre H. Identification of clinical staphylococcal isolates from humans by internal transcribed spacer PCR. *J Clin Microbiol*. 2001;39(9):3099–3103.
17. Pommepuy T, Lons A, Benad K, Beltrand E, Senneville E, Migaud H. Bilateral one-stage revision of infected total hip arthroplasties: report of two cases and management of antibiotic therapy. *Case Rep Orthop*. 2016:3621749.
18. Darrieutort-Laffite C, Andr e V, Leautez S, Tanguy G, Cormier G. Staphylococcus caprae arthritis. *Med Maladies Infect*. 2013;43:131–132.
19. Allignet J, Galdbart JO, Morvan A, et al. Tracking adhesion factors in Staphylococcus caprae strains responsible for human bone infections following implantation of orthopaedic material. *An Microbiol*. 1999;145(8):2033–2042.
20. Blanc V, Picaud J, Legros E, et al. Infection after total hip replacement by Staphylococcus caprae. Case report and review of the literature. *Pathol Biol (Paris)*. 1999;47:409–413.
21. Kawamura Y, Hou XG, Sultana F, et al. Of Staphylococcus species among human clinical specimens and emended description of Staphylococcus caprae. *J Clin Microbiol*. 1998;36(7):2038–2042.
22. Kamme C, Lindberg L. Aerobic and anaerobic bacteria in deep infections after total hip arthroplasty: differential diagnosis between infectious and non-infectious loosening. *Clin Orthop Relat Res*. 1981;154:201–207.
23. Romano CL, Manzi G, Logoluso N, Romano D. Value of debridement and irrigation for the treatment of peri-prosthetic infections. A systematic review. *J Clin Exp Res Hip Pathol Therapy*. 2012;22(S8):19–24.
24. Engesaeter LB, Dale H, Schrama JC, Hallan G, Lie SA. Surgical procedures in the treatment of 784 infected THAs reported to the Norwegian Arthroplasty Register. *Acta Orthop*. 2011;82(5):530–537.

25. Leonard HA, Liddle AD, Burke O, Murray DW, Pandit H. Single- or two-stage revision for infected total hip arthroplasty? A systematic review of the literature. *Clin Orthop Relat Res*. 2014;472(3):1036–1042.
26. Allignet J, Galdbart JO, Morvan A, et al. Tracking adhesion factors in Staphylococcus caprae strains responsible for human bone infections following implantation of orthopaedic material. *An Microbiol*. 1999;145(8): 2033–2042.
27. Allignet J, Aubert S, Dyke KG, El Solh N. Staphylococcus caprae strains carry determinants known to be involved in pathogenicity: a gene encoding an autolysin-binding fibronectin and the ica operon involved in biofilm formation. *Infect Immun*. 2001;69:712–718.
28. Achermann Y, Eigenmann K, Ledergerber B, et al. Factors associated with rifampin resistance in staphylococcal periprosthetic joint infections (PJI): a matched case-control study. *Infection*. 2013;41(2), 431-417.