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CASE REPORT

Yokenella regensburgei OSTEOARTICULAR INFECTION: A CASE REPORT

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ABSTRACT

Yokenella regensburgei belongs to the family Enterobacteriaceae and is an opportunistic agent rarely associated with infections in humans. We report a case of osteoarticular knee infection caused by *Y. regensburgei* in a patient under treatment for rheumatoid arthritis, using corticosteroids, with complication in primary total arthroplasty of the knee. *Y. regensburgei* was identified using the VITEK2 system. Antimicrobial susceptibility testing was performed using the disk-diffusion method, according to the guidelines from the Clinical and Laboratory Standards Institute. The patient presented favorable clinical evolution after the second debridement, with complete removal of the prosthesis and antibiotic therapy with sulfamethoxazole/trimethoprim. This is the first case of *Y. regensburgei* infection described in Brazil.

KEY WORDS: *Yokenella regensburgei*; osteoarticular; infection; sulfamethoxazole; trimethoprim; prosthesis.

INTRODUCTION

Yokenella regensburgei belongs to the family Enterobacteriaceae and is a rarely reported opportunistic human pathogen. Only a few cases have been described worldwide, the majority of which were associated with immunosuppression (Chi et al., 2017; Lo et al., 2011; Wright et al., 2019).

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Due to the small number of reports, the clinical and epidemiological importance of this microorganism has yet to be clarified. It has been isolated from blood, urine, feces, and synovia, as well as from cutaneous wounds and respiratory tract samples (Bhowmick & Weinstein, 2013). Additionally, it has been found in feces of animals that feed on insects, such as bats (Cassel-Béraud & Richard, 1988). It was also described in cases of osteoarticular infections and bacteremia in other countries (Abbott & Janda, 1994). To our knowledge, this is the first reported case of *Y. regensburgei* human infection in Brazil.

CASE REPORT

A 54-year-old female, taking medication for type 2 diabetes and for rheumatoid arthritis (prednisone [20 mg/day], chloroquine [250 mg/day] and ibuprofen [600 mg/day]), presented arthrosis in the left knee. The patient's written consent was obtained. The present study was authorized and approved by Ethics Committee of the Institution (protocol 47937621.7.0000.5082). The HIV test was negative. She underwent primary total arthroplasty of the knee with a bone graft, fixation using two cortical screws, and cementation of the tibial and metallic femoral components. There were no surgical complications, and the patient received antibiotic prophylaxis for 48 hours with cefazolin. On the 40th postoperative day, she developed pain, redness, and a clear exudate from the surgical site, but no fever. After five days, the pain became more intense, and she developed a fistula. Two months after the arthroplasty, the patient was submitted to a new surgery. Extensive synovectomy was performed, and the mobile components of the implant were replaced. A bone fragment was submitted to microbiological culture and *Y. regensburgei* was identified by the VITEK 2 Compact® (bioMérieux) bacterial identification and susceptibility testing system (85% confidence level). The isolate was susceptible to meropenem, tigecycline, and sulfamethoxazole/trimethoprim. Meropenem was used for two weeks. After this initial treatment, the patient felt better with reduced pain and swelling, and oral sulfamethoxazole/trimethoprim therapy was established for four weeks. Two years and seven months later, she presented a new episode of localized disease, with pain lasting for two months and edema in her left knee, followed by redness and function loss of the knee, hindering her gait. Septic loosening of the joint prosthesis was diagnosed. Computer tomography (CT) scan of the left knee demonstrated joint effusion, and a large collection was observed on the medial side of the leg (Figure). Surgical treatment was indicated, with complete removal of the prosthesis. During surgery, purulent secretions and devitalized tissues were observed. Collected material was inoculated in brain heart infusion broth. Grown cultures revealed homogeneous Gram negative rods. The culture was streaked on to MacConkey agar plates resulting in homogeneous morpohology colonies. The bacterium isolated was identified as *Y. regensburgei* by two different systems: VITEK2® and Phoenix® (Becton, Dickinson), with a confidence level in the methods of >95%.

Susceptibility to antimicrobials was evaluated by using the disk-diffusion method, according to the Clinical and Laboratory Standards Institute guidelines (CLSI, 2018). The isolate was susceptible to meropenem, tigecycline, sulfamethoxazole/trimethoprim, ceftazidime, cefepime, amoxicillin/clavulanate, ampicillin/sulbactam, and gentamicin, and resistant to cefazolin. Sulfamethoxazole/trimethoprim intravenous therapy was used, with significant clinical and laboratory recoveries. She was discharged after seven days of intravenous therapy, continuing oral treatment for six months, as chronic osteomyelitis was suspected.

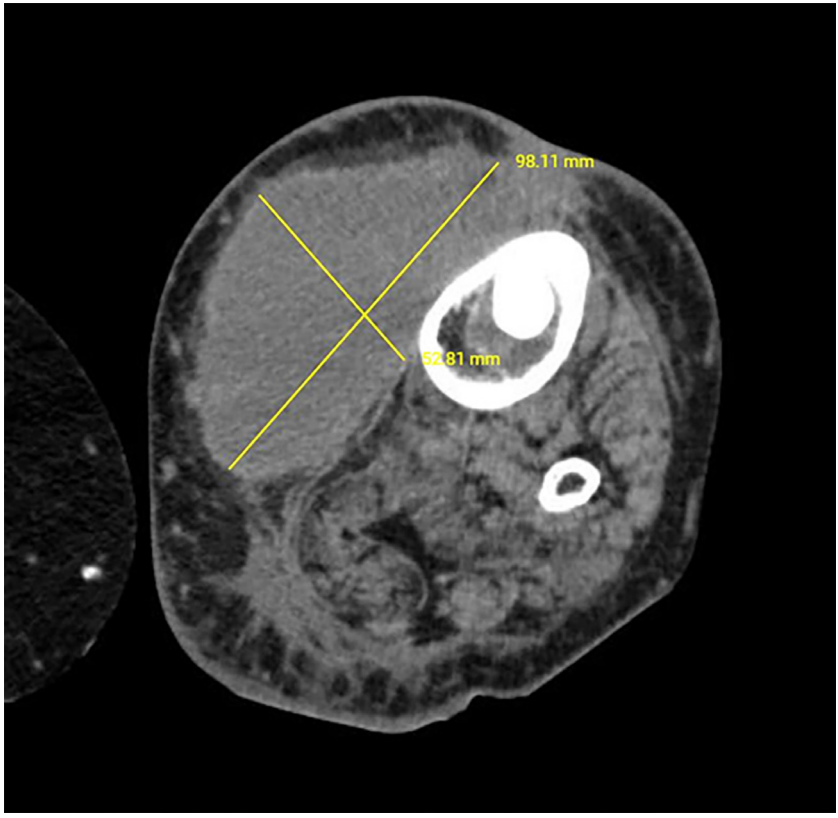

Figure. Computed tomography (CT) scan of the left knee demonstrating joint effusion and large collection. The lines drawn in yellow delimit the anteroposterior diameter of the collection and its transverse diameter, which are the values used for volumetric measurement of the abscess.

DISCUSSION

We describe a rare case of *Y. regensburgei* osteoarticular infection. The patient was immunocompromised due to chronic use of prednisone and chloroquine, presenting type 2 diabetes, comorbidities that have been previously described as predisposing conditions (Lee et al., 2015; Lo et al., 2011). Severe cases of infection by *Y. regensburgei* in immunocompromised patients have also been reported in China and the USA (Bhowmick & Weinstein, 2013; Chi et al., 2017). Cases of *Y. regensburgei* infection in immunocompetent hosts are less common, an example being the case of post-craniotomy osteomyelitis by *Y. regensburgei* in an immunocompetent female patient, who was successfully treated for six weeks with ciprofloxacin and had no recurrence of the infection after one year follow-up (Penagos et al., 2015). Another reported case involved a five-year-old boy who presented a seven-day history of high fever and chills; two of his blood cultures yielded *Y. regensburgei*. This child was treated with ciprofloxacin for seven days with satisfactory clinical response, and no recurrence was detected after a 3 months follow-up (Jain et al., 2013).

One of the limitations of the present study is that we have not performed molecular biology testing to confirm identification of the bacterial species, as there was no immediate access to this resource. However, the species was identified on different occasions and by using two different phenotypic identification systems. Additionally, the antibiotic sensitivity profile observed, also favors *Y. regensburgei* as being the actual pathogen involved, and not *Hafnia* alvei, often mistaken with *Y. regensburgei* even by commercially available identification systems. *H. alvei* exhibits intrinsic resistance to amoxicillin/clavulanate and ampicillin/sulbactam (ANVISA, 2020; Stock et al., 2004), and the disk-diffusion test showed that the pathogen involved in the case described here was susceptible to both antibiotic combinations.

To our knowledge, this is the first report of an infection caused by *Y. regensburgei* in Brazil and illustrates the role of this rare agent in osteoarticular infections.

CONFLICT OF INTEREST

The authors declare no conflicts of interest

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