Chronic Osteomyelitis Due to Tissierella carlieri: First Case

Mona Schweizer,1,4* Guido V. Bloemberg,2,4 Christian Graf,7 Anna L. Falkowski,5,4 Peter Ochsner,6 Peter Graber,1 Sandra Uffler,6 Daniel Goldenberger,1 Vladimira Hinić,7 Susanne Graf,6 and Philip E. Tarr1,5

1Infectious Diseases Service, Kantonsspital Baselland, University of Basel, 2Institute of Medical Microbiology, University of Zurich, Departments of 3Orthopedic Surgery and 4Radiology and Nuclear Medicine, Kantonsspital Baselland, 5Orthopedic Center Brunnmatt, Liestal, 6Microbiology Laboratory, Kantonsspital Baselland, and 7Clinical Microbiology Service, University Hospital Basel, Switzerland

A 54-year-old woman sustained an open fracture of the right femoral shaft after a bicycle accident in 1985. Several operations were performed, the last including internal fixation using a plate and screws. In 2002, acute diverticulitis was treated with ciprofloxacin/metronidazole. In 2007, she presented with pain and swelling of the right thigh. Plain radiographs showed radiolucent femoral areas (Figure 1a and b). Treatment was with debridement and removal of orthopedic hardware except for a difficult-to-remove screw fragment. Gentamicin beads were temporarily placed along the femur. Eight intraoperative specimens showed no growth after 10 days. Because of poor wound healing, a computed tomography scan was obtained, showing a sinus tract, femoral cortical defects, and intramedullary air (Figure 1c–e). Additional debridement was done; cultures of intraoperative biopsy specimens again remained sterile. Amoxicillin/clavulanate was prescribed for 2 weeks, and ciprofloxacin/rifampin was prescribed for 10 weeks.

In 2008, local pain and swelling reappeared. Magnetic resonance imaging showed chronic osteomyelitis, contiguous with the screw fragment. This was removed, and the medullary cavity was reamed from proximal to distal [1]. Seven intraoperative specimens showed no growth after 10 days. Amoxicillin/clavulanate was prescribed for 3 months. In 2010, local swelling reappeared. Surgical bone biopsies showed no growth at 10 days. Bacterial broad-range 16S rRNA gene polymerase chain reaction (PCR) was done, and sequence analysis showed a Tissierella species. At 3 weeks, 3 of 7 biopsies showed pure anaerobic growth of a Tissierella species in brain-heart-infusion broth. The isolate was positive for arginine dihydrolase, alkaline...
phosphatase, and pyroglutamic acid arylamidase (rapid ID32A test strip; bioMérieux, Switzerland) and leucine arylamidase, tryrosine arylamidase, phenylalanine arylamidase, and pyroglutamic acid arylamidase (VITEK, bioMérieux, Switzerland). Identification was done by sequencing almost the entire 16S rRNA gene. The 1434 base pair (bp) sequence deposited in GenBank (accession no. KR781503) showed 1 mismatch over a 1371 bp segment compared with reference *Tissierella carlieri* strain LBN 292 (99.9% sequence similarity) that was recently described [2]. Susceptibility testing results are listed in Table 1.

No antimicrobial therapy was started because the pathogenic role of the *Tissierella* was unclear, and the wound had healed. In November 2011, local pain and discharge from the scar were noted. Imaging revealed osteomyelitis (Figure 2), including an area with sequestra in the lateral femoral condyle, an area not debrided until then. In January 2012, the distal three fifths of the medullary cavity were reached by a long cortical window from lateral. Intraoperatively, a sinus tract and several bony sequestra were found. Extensive intramedullary debridement was performed, and a cement spacer containing gentamicin/clindamycin/meropenem beads was temporarily placed. Aerobic and anaerobic cultures from 16 surgical specimens remained sterile after 6 weeks; broad-spectrum PCR again showed the isolate might have been selected for by previous, prolonged antimicrobial treatments in the setting of persistent dead bone [7–9]. Debridement of dead bone is a key feature of successful management [11–13]. Our case also suggests that, in culture-negative osteomyelitis cases, attention should be paid to careful specimen collection and prolonged anaerobic culture. Microbiological diagnosis may require 16S rRNA gene sequence analysis based on broad-range PCR. These results can then be used to guide optimal culture method [14].

**CONCLUSIONS**

Most cases of anaerobic osteomyelitis have been polymicrobial [7–10]. In our patient, monomicrobial infection with a *T carlieri* isolate might have been selected for by previous, prolonged antimicrobial treatments in the setting of persistent dead bone [7–9]. Debridement of dead bone is a key feature of successful management [11–13]. Our case also suggests that, in culture-negative osteomyelitis cases, attention should be paid to careful specimen collection and prolonged anaerobic culture. Microbiological diagnosis may require 16S rRNA gene sequence analysis based on broad-range PCR. These results can then be used to guide optimal culture method [14].

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