**Abstract**

We report a case of an 88-year-old man with osteomyelitis of the right ankle, with histopathology demonstrating a *Mycobacterium* spindle cell pseudotumor. The *Mycobacterium* contained in this spindle cell pseudotumor was *Mycobacterium chelonae*. *M. chelonae* spindle cell pseudotumors are rare and have only been reported twice previously in the literature. Similarly, *M. chelonae* presenting as the pathogen in bone infection is rare. Due to this unusual presentation of *M. chelonae*, the antibiotic rationale was based largely on case reports and consisted of imipenem, clarithromycin, and linezolid. Antibiotic complications were experienced by the patient. Despite a renally adjusted dose of imipenem, the patient experienced imipenem toxicity and his antibiotics were modified to tigecycline and clarithromycin. Although his symptoms were clinically resolving, the patient sadly passed away before completing treatment.

**Keywords:** *Mycobacterium chelonae*, osteomyelitis, spindle cell pseudotumor

**INTRODUCTION**

The presentation of *Mycobacterium chelonae* is unusual in the setting of bone infection[1–4] and the presentation of *M. chelonae* as a spindle cell pseudotumor even more so.[5] The setting of this unusual case presentation is documented alongside the antibiotic therapy used and the rationale behind this. It highlights complications to this treatment, namely imipenem toxicity, despite renal dose adjustment.

**CASE REPORT**

An 88-year-old man presented with a 6-week history of pain in his right ankle and erythema over the right lower leg. The lateral malleolus and lateral part of the calcaneum were swollen and tender, and he was unable to weight-bear on the right foot. He also reported a history of weight loss, change in bowel habit, and loss of appetite over the previous 6 months. He had an extensive medical history: chest wall melanoma excised the year before; atrial fibrillation; type 2 diabetes mellitus; gout; chronic kidney disease; a coronary artery bypass graft insertion; and right total knee replacement. The patient also reported an allergy to penicillin.

A magnetic resonance imaging of the right foot showed a distal fibular lytic lesion, suggestive of osteomyelitis, extensive cellulitis with peroneal tenosynovitis, and a calcaneal lesion with features of a bone infarct.

A bone biopsy of the right ankle initially reported acid-fast bacilli, with histology showing evidence of a mycobacterial spindle cell pseudotumor [Figures 1 and 2]. The bone biopsy sample demonstrated rapid mycobacterial growth and was referred to the Mycobacterial Reference Laboratory, where the isolate was identified as *M. chelonae* (through polymerase chain reaction-based rapid identification).[6]

*M. chelonae* is typically associated with immunocompromise, and computed tomography of the chest, abdomen, and pelvis was performed. This found an appendix mucocele and new lung nodules in the right lung but no definitive evidence of malignancy.

The initial empirical choice of antibiotics was imipenem 250 mg three times a day (a renally adjusted dose due to imipenem toxicity). Despite a renally adjusted dose of imipenem, the patient experienced imipenem toxicity and his antibiotics were modified to tigecycline and clarithromycin. Although his symptoms were clinically resolving, the patient sadly passed away before completing treatment.
the patient’s estimated glomerular filtration rate of 35), clarithromycin 500 mg twice a day, and co-trimoxazole 960 mg twice a day. Imipenem is the preferred carbapenem in *M. chelonae* infection.[2]

Following receipt of the Reference Laboratory sensitivities (reported sensitivity to amikacin, clarithromycin, and linezolid with resistance to cefoxitin, co-trimoxazole, ciprofloxacin, moxifloxacin, and doxycycline), the patient was switched to imipenem, clarithromycin, and linezolid. Previous studies have found that clarithromycin in combination with linezolid is clinically effective in immunocompromised patients with cutaneous *M. chelonae*.[7] The patient was sent home with outpatient antimicrobial therapy and regular blood monitoring.

Several complications were encountered with this choice. Most significantly, despite a renally adjusted dose of imipenem, the patient was re-admitted 12 days after starting treatment with confusion. Mild diffuse encephalopathy was diagnosed on electroencephalogram but no seizure activity. These symptoms did not reoccur after imipenem was stopped. Second, the patient began to experience myelosuppression secondary to linezolid (after 12 days of treatment).

Following his readmission, the patient was switched to a combination of tigecycline and clarithromycin. Tigecycline has been reported as improving outcomes as part of a multidrug regimen to treat *M. chelonae*.[8]

There is no firm evidence upon which to base the duration of treatment in cases of osteomyelitic *M. chelonae*. Previous case reports have documented mycobacterial spindle cell pseudotumor caused by *Mycobacterium intracellulare* that took 2 years of antibiotic treatment to resolve.[9]

The literature suggests that along with antibiotics, surgical intervention is vital to treating *M. chelonae* bone infection. However, given the patient’s extensive medical history, he was not believed to be fit for surgery, and therefore, debridement of the bone was not performed.

Clinically, the inflammation of the ankle had begun to subside before readmission, and the patient’s symptoms continued to improve with tigecycline and clarithromycin [Figure 3] although the patient’s inflammatory markers remained static.

After 16 days on this combination of antibiotics, the patient became hypotensive and tachycardic and sadly passed away, with the cause of death found to be a combination of restrictive cardiomyopathy and multiorgan failure. The exact nature of the underlying immunosuppressive pathology remains unknown.

**DISCUSSION**

Mycobacterial spindle cell pseudotumors are benign lesions that present histologically as spindle-shaped histiocytes containing acid-fast bacilli. They are most commonly caused by *Mycobacterium avium* and *Mycobacterium tuberculosis* and usually present in skin, lungs, eyes, and soft tissue.[1] The case...
presented here is unusual in two respects. The *Mycobacterium* spindle cell pseudotumor was formed by *M. chelonae*, a finding that has only been reported twice previously in the literature.\(^5\)

Second, *M. chelonae* presenting in bone has only been reported in nine cases in the literature.\(^{[1‑3]}\)

Spindle cell pseudotumors are almost always associated with immunodeficiency: a review of *Mycobacterium* pseudotumor cases found that of the 23 cases of *Mycobacterium* spindle cell pseudotumors in the literature, 21 patients had immunosuppression either as a result of HIV/AIDS, immunosuppressive therapy, or diabetes.\(^5\)

Given the patient’s history of melanoma, bone metastases were among the initial differential diagnoses. The literature reports that spindle cell pseudotumors are often mistakenly diagnosed radiologically as malignant lesions, for example, sarcomas, schwannomas or histiocytomas, until biopsy confirms otherwise.\(^9\)

Carbapenems are known to lower seizure thresholds and can cause confusion. Imipenem is primarily renally excreted, and therefore, dose adjustment in kidney failure is recommended to limit side effects. In this case, however, side effects occurred despite renal adjustment of the imipenem dose, which resolved on discontinuation of the treatment.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

**REFERENCES**

1. Talanow R, Vieweg H, Andresen R. Atypical osteomyelitis caused by *Mycobacterium chelonae*-A multimodal imaging approach. Case Rep Infect Dis 2013;2013:528795.
2. Rahman I, Bhatt H, Chilag S, Duffus W. *Mycobacterium chelonae* vertebral osteomyelitis. South Med J 2009;102:1167-9.
3. Oelberg DA, Mendelson J, Miller MA, Dascal A. Disseminated *Mycobacterium chelonae* infection presenting as progressive multifocal osteomyelitis: Report of two cases and a review of the literature. Can J Infect Dis 1994;5:28-32.
4. Al-Knawy M, Bosch W, Garner H, Miraecidi M, Cawley J, Murray P, et al. A descriptive analysis of nontuberculous mycobacterial infections (NTM) of the upper extremity. Int J Mycobacteriol 2015;4:37-8.
5. Sfeir M, Soave R, Van Besien K, Satlin M, Jenkins S, Westblade L, et al. Mycobacterial spindle cell pseudotumor: Epidemiology and clinical outcomes. Open Forum Infect Dis 2016;3:563.
6. Brown-Elliott BA, Philley JV. Rapidly growing mycobacteria. Microbiol Spectr 2017;5:703-4.
7. Parize P, Hamelin A, Veziris N, Morand PC, Guillemain R, Lortholary O, et al. Induction therapy with linezolid/clarithromycin combination for *Mycobacterium chelonae* skin infections in immunocompromised hosts. J Eur Acad Dermatol Venereol 2016;30:101-5.
8. Wallace RJ Jr., Dukart G, Brown-Elliott BA, Griffith DE, Scepella EG, Marshall B, et al. Clinical experience in 52 patients with tigecycline-containing regimens for salvage treatment of *Mycobacterium abscessus* and *Mycobacterium chelonae* infections. J Antimicrob Chemother 2014;69:1945-53.
9. Ismail I, Carey M, Trotter S, Kunst H. Mycobacterial spindle cell pseudotumour of the brain in a patient with sarcoidosis. BMJ Case Rep 2015;2015. pii: bcr2014206171.