

CAS CLINIQUE / CASE REPORT

OSTEOMYELITIS AT TWO NONCONTIGUOUS SITES CAUSED BY *MYCOBACTERIUM MARINUM* IN AN IMMUNOCOMPETENT HOST

Case Report and Literature Review

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ABSTRACT : Nontuberculous mycobacteria are rare causes of skin, soft tissue, and musculoskeletal infections. *Mycobacterium marinum* remains one of the most commonly encountered mycobacterial species in humans, causing superficial cutaneous as well as deep infections. We are reporting a case of *M. marinum* osteomyelitis involving two primary noncontiguous sites in an immunocompetent host, which was successfully treated with surgical drainage and antibiotic therapy.

Keywords : *Mycobacterium marinum*, osteomyelitis, cellulitis, immunocompetent

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RÉSUMÉ : Les mycobactéries autres que tuberculeuses sont de rares causes d'infections de la peau, des tissus mous ou d'infections locomotrices. *Mycobacterium marinum* reste l'espèce la plus rencontrée chez l'homme. Elle provoque des infections cutanées superficielles ou profondes. Nous rapportons un cas d'ostéomyélite à *M. marinum* qui a affecté deux sites non contigus chez un patient immunocompétent traité avec succès par drainage chirurgical et antibiothérapie.

Mots-clés : *Mycobacterium marinum*, ostéomyélite, cellulite, immunocompétent

INTRODUCTION

Nontuberculous mycobacteria (NTM) are rare etiologic agents of osteomyelitis and have been implicated in skin, soft tissue, joint, and bone infections [1]. Individuals with immunodeficiency, such as HIV infection or malignancy, are more prone to develop NTM osteomyelitis [1-3]. However, few cases of NTM osteomyelitis in immunocompetent hosts have been reported in the literature [4-9].

Bacterial pathogens, particularly Gram-positive cocci, remain the most common causes of skin and bone infections. Among atypical mycobacteria, *Mycobacterium marinum* is the most frequently encountered species causing cutaneous infections in humans but is a rare cause of osteomyelitis [10-11]. *M. marinum* can cause superficial cutaneous as well as deep infections involving tendons and small bones particularly in the hands and feet [11-12]. Infection by *M. marinum* usually occurs when a traumatized skin surface comes in contact with contaminated water [7, 10-12]. We are describing an immunocompetent patient with *M. marinum* osteomyelitis involving two primary noncontiguous sites, which was successfully treated with surgical drainage and antibiotic therapy.

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CASE REPORT

A 62-year-old white male US citizen presented to the American University of Beirut (AUB) Medical Center with pain, redness, and swelling of his right foot. He is a college professor and resides in the faculty apartments of the AUB campus. Two weeks prior to presentation, he had a superficial wound at the site of a ligament tear in the lateral aspect of his right foot as he was walking on the beach. He was initially treated with amoxicillin-clavulanate and clindamycin for suspected cellulitis without substantial improvement in his clinical condition. One week after initiation of antibiotic treatment, he presented to the emergency room. He denied any history of fever, chills, or night sweats. Upon examination, the patient was afebrile. An area of cellulitis was noted over the lateral aspect of his right foot, with a purulent crater at the center but no frank drainage (Figure 1a). A plain radiographic film was suspicious for osteomyelitis. The patient was admitted and started on levofloxacin to cover both Gram-positive and Gram-negative pathogens after obtaining a superficial wound swab culture. The next day, swelling and mild redness were noticed over the right fifth metacarpophalangeal joint at the site of a recent blunt trauma (Figure 1b). Upon further questioning, the patient admitted to having soaked his right hand and foot in icy tap water over the few days preceding the onset of his symptoms, with the same water and basin being reused several times.

Basic hematologic and biochemistry laboratory tests were normal except for mild anemia (Hb and Hct were

10.9 g/dL and 33% respectively). Inflammatory markers were within normal limits (C-reactive protein: 0.8 mg/L and erythrocyte sedimentation rate: 5 mm/min at one hour).

Magnetic resonance imaging of the right hand and foot revealed osteomyelitis of the fifth metacarpal joint with swelling and abscess formation, and destruction of the fifth metatarsal bone with diffuse surrounding edema compatible with osteomyelitis, deep cellulitis with small abscesses, and septic arthritis of the fifth metatarsophalangeal joint (Figure 2 a & b). A bone scan was then ordered to determine whether additional sites were involved and confirmed the presence of osteomyelitis at the right fifth metacarpal and right fifth metatarsal bones only. Superficial wound cultures from the foot grew *Citrobacter freundii*, *Enterobacter cloacae* and *Pseudomonas aeruginosa*.

The patient was shifted to ciprofloxacin 600 mg IV every 12 hrs. Blood cultures and HIV antibodies were negative. Despite the absence of fever and positive blood cultures, a transesophageal echocardiogram was performed to rule out any evidence of systemic dissemination and did not show evidence of endocarditis. At that stage, because of the lack of improvement with antibacterial agents, and because of the history of water exposure, mycobacterial infection was suspected. On day 5 of admission, the patient underwent incision and drainage with sequestrectomy of the right fifth metacarpal and right fifth metatarsal bones. Tissue samples were sent for microbiological and histological examination.

Tissue pathology revealed chronic osteomyelitis with necrotizing granulomatous inflammation. The acid-fast stain was negative. Cultures on Löwenstein-Jensen medium at 30 °C showed cream-colored photochromogenic colonies that turned yellow upon exposure to light. The organism was subsequently identified as *M. marinum* based on biochemical characteristics. The patient was treated with a 6-month course of clarithromycin and

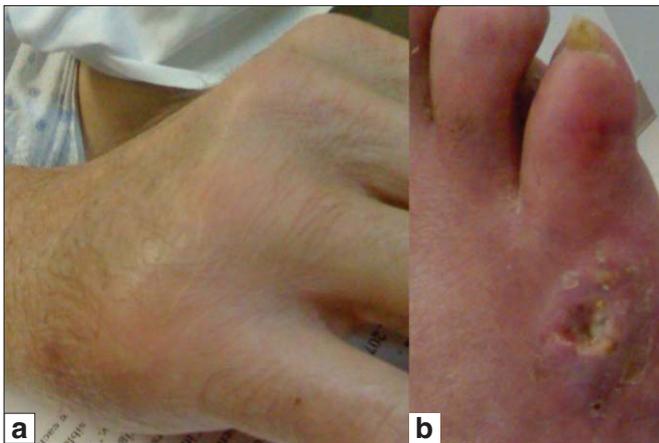


FIGURE 1. Photograph of the patient's hand and foot showing swelling and erythema over the base of the right fifth finger (a), and a necrotic lesion at the base of the right fifth toe surrounded by erythema (b).

trimethoprim/sulfamethoxazole with successful results. At one year follow-up, the patient demonstrated good healing at the site of surgery and resolution of all symptoms with restoration of good function.

DISCUSSION

Our patient was found to have *M. marinum* osteomyelitis at two primary noncontiguous sites in the hand and foot. One hypothesis would be that the infection was acquired upon repeated exposure to contaminated tap water that was re-used several times. We attempted to confirm our hypothesis by doing mycobacterial cultures on the tap water from the patient's residence, which were negative. Unfortunately, the basin with the used water was no longer available and we could not prove a causal relationship. Another hypothesis is infection of the foot with *M. marinum* upon exposure to sea water that subsequently contaminated the water basin used to soak both the foot and the hand. Although mycobacterial cultures on blood were not performed, hematogenous spread was deemed less likely given the negative bone scan and echocardiogram, and the exclusive involvement of areas exposed to the same suspected source.

Only two cases of osteomyelitis with *M. marinum* involving two noncontiguous sites have been described in immunosuppressed individuals. The first patient was a child with severe combined immunodeficiency who



FIGURE 2. Magnetic resonance imaging of the patient's hand and foot showing osteomyelitis and destruction of the fifth metacarpal bone with surrounding soft tissue swelling and abscess formation (a), and osteomyelitis and destruction of the fifth metatarsal bone with septic arthritis of the fifth metatarsophalangeal joint, deep cellulitis, and formation of small abscesses (b).

TABLE I
PUBLISHED CASES of *MYCOBACTERIUM MARINUM* OSTEOMYELITIS in IMMUNOCOMPETENT PATIENTS

AUTHORS [Reference]	Year	Sex	Type of exposure	Site	Clinical presentation
WILLIAMS <i>et al.</i> [9]	1970	Male	Steel cable	Hand	Localized infection
WILLIAMS <i>et al.</i> [9]	1970	Male	Not identified	Hand	Extensive localized infection
WENDT <i>et al.</i> [8]	1986	Female	Not identified	Hand	Localized infection
CLARK <i>et al.</i> [4]	1990	Male	Fisherman	Hand	Localized infection
VAZQUEZ <i>et al.</i> [7]	1992	Female	Fish tank	Hand	Localized osteomyelitis; diffuse cutaneous lesions
HARTH <i>et al.</i> [5]	1994	Male	Fish tank	Hand	Localized infection
SHIH <i>et al.</i> [6]	1997	Female	Fish dealer	Hand	Localized infection
PRESENT CASE	2009	Male	Tap water or sea water	Hand + foot	Two sites of osteomyelitis

presented with diffuse cutaneous lesions and osteomyelitis of the hand and foot in the setting of bacteremia [13]. The second patient was a 48-year-old woman with Jo-1 syndrome on cyclophosphamide and prednisone that presented with osteomyelitis of the hand and foot along with polyarthritis and subcutaneous nodules [14]. To our knowledge, and based on literature searches on PubMed, no cases of *M. marinum* osteomyelitis involving two primary sites have been reported so far in an immunocompetent patient (Table I).

Soft tissue and musculoskeletal infections with *M. marinum* are usually acquired when injured skin is exposed to contaminated salt or fresh water [12]. Three types of possible exposure are described in the literature: contact with contaminated aquarium water, fish- or shellfish-associated injuries, and salt water-associated injuries [10-12]. *M. marinum* skin infections can be clinically classified into three types: type I consisting of self-limited, superficial, ulcerated or crusted lesions; type II consisting of single or multiple subcutaneous granulomas, with or without ulcerations; and type III consisting of deep infections involving tissues like tendons, joints, and bones [9], as was the case with the patient we are describing.

The treatment of deep-seated *M. marinum* infections can be difficult and often requires the use of combination therapy [15]. While superficial infections usually respond to antibiotic therapy alone [12] and sometimes resolve spontaneously [4], deep infections often require surgical debridement of necrotic tissue in addition to antibiotic therapy [12]. The organism is inherently resistant to isoniazid and pyrazinamide and intermediately susceptible to streptomycin. Antibiotic choices include rifampin, ethambutol, clarithromycin, tetracyclines, and trimethoprim/sulfamethoxazole [4]. Our patient showed a progressive and sustainable response to a combination of clarithromycin and trimethoprim/sulfamethoxazole.

We conclude that clinicians should have a high index of suspicion for *M. marinum* infection in immunocompetent as well as immunodeficient patients with a history of contact with contaminated water.

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