

Mycobacterial Tenosynovitis After Sea Urchin Spine Injury in an Immunocompromised Patient

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CASE PRESENTATION

A 46-year-old, right-hand dominant, female patient presented with progressive pain and swelling of the left hand associated with multiple small, raised nodules over the volar index finger and thumb extending to the palm. The patient reported having accidentally placed her left hand on a sea urchin while diving in Hawaii 1 month before evaluation. She immediately experienced pain and noted multiple sea urchin spines had punctured her volar left index finger, thumb, and palm. The patient presented to a local medical facility where she received tetanus vaccination and was prescribed 1 week of oral cephalexin. After returning home, the pain and swelling progressed, prompting further evaluation in the emergency department on 2 more occasions. An additional 3-day course of doxycycline and cephalexin was given, and superficial bedside debridement of the nodules in the emergency department was performed; however, symptoms continued to worsen. The patient was ultimately referred to hand surgery clinic.

The patient had a history of metastatic ovarian cancer and had been receiving carboplatin and gemcitabine chemotherapy, complicated by pancytopenia. Her medical history was also notable for Crohn's disease on azathioprine, renal vein

thrombosis on therapeutic anticoagulation, and methylenetetrahydrofolate reductase mutation. On initial evaluation 1 month after her injury, she denied any recent fever and attributed mild nausea, weakness, fatigue, and malaise to chemotherapy. Her left index finger and thumb were held in 20 degrees of interphalangeal joint flexion with associated fusiform edema (Figure 1). She had significant pain on palpation of the flexor tendon sheath and pain with passive extension. Multiple small, firm, slightly pale, tender nodules were noted over the volar index finger and thumb extending to the distal palm. An x-ray evaluation of the left hand noted only soft tissue swelling. Ultrasound demonstrated focal areas of linear echogenicity in the index finger and thumb with associated surrounding fluid.

The patient was admitted to the hospital, and the left index finger and thumb were explored in the operating room. Palpable subcutaneous lesions associated with each nodule were excised and found to be less than 1 mm in diameter. Significant inflammation and edema were noted. The flexor tendon sheath was distended and upon entry, and there was immediate drainage of pale, gelatinous material (Figure 2). Marked synovitis was noted. Specimens were sent for aerobic, anaerobic, mycobacterial, and fungal cultures as well as histologic review. A1 pulley release was performed with synovectomy of the involved index finger and thumb. The patient's postoperative course was uneventful, and she was discharged the following day on oral ciprofloxacin after a 1-time dose of 1.5 g intravenous dalbavancin.

Routine bacterial cultures and acid-fast smear were negative. Pathologic review of excised nodules revealed foreign body reaction. Mycobacterial cultures obtained in liquid media (BACTEC/ALERT) from synovial tissue and from subcutaneous nodules both ultimately grew *Mycobacterium chelonae* after 4 weeks, identified at the University of Texas Health Science Center by *rpoB* sequencing. The patient was treated with 400 mg of moxifloxacin (oral) daily and 500 mg of azithromycin daily for an anticipated 6-month course. Susceptibility testing (BACTEC/ALERT) occurred 1 month postoperatively and demonstrated susceptibility to linezolid (minimum inhibitory concentration [MIC] 4 mg/L), amikacin (16 mg/L), tobramycin (<1 mg/L), doxycycline and minocycline (<1 mg/L), and clarithromycin. The tigecycline MIC was 0.25 mg/L. Furthermore, the isolate was intermediate to ciprofloxacin and moxifloxacin (2 mg/L) and imipenem (8 mg/L) and resistant to trimethoprim-sulfamethoxazole and cefoxitin. The patient's antibiotic regimen was switched to oral doxycycline 100 mg daily and azithromycin 500 mg daily for 1 month, but the patient did not tolerate doxycycline due to significant gastric reflux symptoms. After discussion with the patient's psychiatrist regarding concomitant use of sertraline, she was started on 600 mg of linezolid twice daily

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Figure 1. Preoperative examination was notable for fusiform edema of the index finger and thumb, held in slight flexion, with multiple pale, tender, subcutaneous nodules over the volar aspects.

in addition to azithromycin with a decrease in sertraline dose and close monitoring for evidence of serotonin syndrome and thrombocytopenia [1]. Her wounds healed well with improvement in pain, edema, and hand function. She resumed chemotherapy on postoperative week 3. Azathioprine was not held during treatment. Approximately 1 month later, given progression of her cancer, her chemotherapy was changed to docetaxel. At 10 weeks follow up, her hand function was normal. Her subsequent long-term antibiotic course is being determined in an ongoing fashion due to progression of oncologic disease.

Human and Animal Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Informed Consent

Informed consent was obtained from the individual participant included in this article.



Figure 2. Intraoperatively, the flexor tendon sheath was opened to reveal marked synovitis with pale, gelatinous appearance.

DISCUSSION

Our case represents the first report of *Mycobacterium chelonae* isolated from a sea urchin-related granuloma. Mycobacterial infections associated with exposure to sea water have been reported occasionally in the literature, most often caused by *Mycobacterium marinum*, in association with trauma related to fishing injuries or abrasions [2–6]. There is 1 case report of an immunocompetent patient who developed *M marinum* osteomyelitis of the first metatarsal after stepping on a sea urchin [7]. De la Torre et al [8] examined 50 pathologic specimens of sea urchin granulomas and found granulomatous features suggesting additional mycobacterial infection. Using polymerase chain reaction amplification of a 924-base pair deoxyribonucleic acid fragment from mycobacterial 16S ribosomal ribonucleic acid genes, 8 positive biopsy specimens from 7 patients were identified (21%). *Mycobacterium marinum* restriction patterns were positive in 3 of the 8 samples [8].

Mycobacterial tenosynovitis of the hand has been specifically reported in the literature, usually associated with puncture wounds and exposure to sea water and fish [9–12]. Due to the indolent nature of mycobacterial infection, onset of symptoms is typically gradual and delay in diagnosis and surgical treatment, often by months, is common [13]. Retention of spine fragments is known to cause painful skin nodules with foreign-body granulomatous reaction as well as arthritis and synovitis [14]. Diagnosis of mycobacterial tenosynovitis requires culture of the involved synovial tissue; however, initial smear and stains are often negative, as was the case in our patient. Identification of retained sea urchin spines may be suggested radiographically by x-ray, ultrasound, or magnetic resonance imaging [15], but small fragments associated with cutaneous nodules may not be apparent. Histologic evaluation typically demonstrates granulomatous inflammatory reaction [10, 16].

The recommended initial treatment for suspected mycobacterial tenosynovitis is operative exploration with limited synovectomy [16]. Treatment of painful skin nodules after sea urchin spine injury entails removal of foreign bodies and the associated granulomas resulting in improvement in pain, resolution of systemic symptoms, and fewer long-term complications [17]. Surgical excision of foreign bodies and debridement of infected tissue is therefore considered essential to recovery from *M chelonae* skin and soft tissue infection [18]. After identification of the mycobacterial pathogen, consultation with the infectious disease section is recommended for 6 to 12 months of multidrug antimycobacterial therapy [19]. Drug-susceptibility testing should be performed if laboratory capabilities permit, given previously described low rates of susceptibility to oral agents [18]. Antimicrobial selection for treatment of *M marinum* infection has been reviewed, and the mainstay of therapy remains macrolides, tetracyclines, and fluoroquinolones, alone or in combination [20].

CONCLUSIONS

The diagnosis and treatment of atypical mycobacterial tenosynovitis requires a high index of suspicion based upon a history of penetrating injury to the hand, exposure to sea water or fish, and typically indolent clinical course. In this case, the patient's symptoms were not responsive to oral antibiotics alone and progressed more quickly than might be expected if she had normal immune function. Treatment with operative exploration, synovectomy, and subcutaneous foreign-body granuloma excision, followed by multidrug oral therapy was successful in this case.

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