An Unexpected Cause of Tenosynovitis

To the Editor:

Legionella species are significant pathogens with most infections being caused by Legionella pneumophila and presenting as Legionnaire disease or Pontiac fever. Extrapulmonary legionellosis is uncommon and is often thought to arise from hematogenous spread after primary pulmonary infection. To date, the extrapulmonary sites of infections that have been reported in the literature include spleen, kidneys, liver, infective endocarditis, myositis, cellulitis, and cutaneous abscesses. The majority of extrapulmonary Legionella infections have been described in immuno-compromised patients.

A 69-year-old female florist presented to hospital with 1 week of worsening pain and swelling of the right hand, having been treated in the community for presumed seronegative arthritis for the preceding 6 months. Her presentation 6 months earlier was with pain and swelling in her right hand and mild, transient symptoms in the left hand. Clinical examination was normal. The tenosynovitis was confirmed on ultrasound (US). She was treated with ceftriaxone, but failed to improve. Synovial aspirate was not performed, but she had negative blood cultures. She commenced treatment with methotrexate 10 mg/week and hydroxychloroquine 400 mg/day. However, she had an incomplete response. The flexor tendon compartment was injected with corticosteroids 1 month prior to the current hospital admission with temporary improvement in her symptoms. She did not respond to a 4-day course of prednisolone (50 mg/day) immediately prior to the hospital presentation. There were no erosive changes (Figure 1). Radiographs of her left hand, both feet, and chest were normal. US of the right hand and wrist demonstrated flexor tenosynovitis, carpal, and fourth MCP effusions. A synovial fluid aspirate from the right wrist cultured Staphylococcus warneri with a leukocyte count of 0, which was thought to be a contaminant. There were no crystals.

Magnetic resonance imaging demonstrated extensive subcutaneous edema throughout the right hand with florid tenosynovitis of the flexor tendons and a fluid collection deep to the flexor tendon compartment (Figures 2A and 2B). A washout and synovial biopsy were performed. Synovial tissue histopathology demonstrated active chronic inflammation and no organisms were seen. Initial bacterial (pediatric BACTEC culture), mycobacterial, and fungal cultures of both the synovial tissue and fluid were negative. The synovial tissue was set up on buffered charcoal yeast extract agar and typical Legionella longbeachae colonies were identified on Day 5 by MALDI-TOF (Matrix-Assisted Laser Desorption Ionization Time of Flight Mass Spectrometry; Bruker Corp.). The L. longbeachae was unlikely to be a contaminant given the clinical scenario and her potential occupational exposure. She was prescribed a 6-week course of 750 mg BID ciprofloxacin. She responded well and has regained full hand function with no recurrence of her joint symptoms 6 months after completing the antibiotic course.

L. longbeachae was initially isolated in Long Beach, California, in 1981 and has subsequently been linked to potting mixes. Poor hand hygiene after gardening and exposure to aerosolized organisms have been linked to infection through inhalation or ingestion. To our knowledge, ours is the first reported case of L. longbeachae tenosynovitis without preceding injury or clinical pulmonary infection. There has been 1 published case report of septic arthritis in an immunosuppressed patient with rheumatoid arthritis. There have been other reports of soft tissue and musculoskeletal infections with L. longbeachae in the context of concomitant pneumonia or direct inoculation through trauma. Rare subtypes, such as L. bozemanae, have also been reported to cause septic arthritis.

Legionella infections are often underdiagnosed, especially in extrapulmonary sites. This case highlights the importance of considering rare infective etiologies in the differential diagnosis of subacute or chronic unilateral tenosynovitis, especially in immunocompromised hosts, those with potential occupational exposures, or those not responding to treatment as expected.

LOUISA CHOUST*, MBBS(Hons), Rheumatology Department, Royal Melbourne Hospital, Melbourne, Australia; JESSICA DAY*, MBBS(Hons), Rheumatology Department, Royal Melbourne Hospital, Melbourne, Australia; SABINA CICIRIELLO, MBBS, PhD, Rheumatology Department, Royal Melbourne Hospital, Melbourne, Australia. * These authors contributed equally to this work.

REFERENCES


3. www.jrheum.org
Figure 2. (A and B) Magnetic resonance imaging of the right hand demonstrating extensive subcutaneous edema throughout the hand with florid tenosynovitis of the flexor tendons and a fluid collection deep to the flexor tendon compartment.


J Rheumatol 2016;43:2; doi:10.3899/jrheum.150968