

# The sequelae of late diagnosis in tuberculous flexor tenosynovitis of the hand—a report of 2 cases

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## Case 1

A 57-year-old immigrant worker from former Yugoslavia presented with a 10-month history of painful and recurrent swelling of the flexor tendon sheaths of the left wrist with severely limited active digital flexion. He had received non-steroidal anti-inflammatory drugs and physical therapy with no beneficial effects. Laboratory findings included an elevated ESR, and a positive tuberculin skin test reaction. The patient had no history of tuberculosis, but the chest radiograph revealed scar tissue in the left upper lobe, which was suggestive of old, healed tuberculosis. Plain radiographs of the left hand were normal, except for a slight soft tissue swelling on the palmar aspect of the wrist.

At surgery, we found a compressed median nerve in the carpal tunnel, synovial fluid, and a caseous mass within the wall of the flexor tendon sheaths. The superficial and deep flexor tendons were partially ruptured and adhered to each other (Figure 1). Tenosynovectomy with complete excision of the affected tissues was performed and the median nerve was released.

Tuberculous tenosynovitis was confirmed by histological examination of the synovial exudate, the caseous mass showing necrosis, acute inflammation, and numerous caseating granulomas, as well as by cultures positive for *Mycobacterium tuberculosis*. Gentle mobilization was started 4 days after surgery. Postoperative management, including administration of pyrazinamide, rifampin, and isoniazid for 3 months, was started after consultation with the internist. Drug susceptibility tests, confirming sensitivities to all anti-tuberculous drugs, were completed after chemotherapy was initiated. The use of rifampin and isoniazid was continued for an additional 9 months to ensure adequate treatment of this advanced stage of the disease.

Pain and the swelling resolved rapidly. Follow-up at 4 years showed no signs of recurrence and normal wrist radiographs. The patient regained useful wrist motion, but he remained unable to flex his fingers. He was satisfied with the outcome and declined further surgical reconstruction.

## Case 2

A 70-year-old Austrian woman was admitted for evaluation of intermittent pain and swelling on the dorsoradial aspect of the right hand. She had been treated for recurrent flexor tenosynovitis of the wrist, and subsequent mild reflex sympathetic dys-



Figure 1. Case 1. Intraoperative photograph showing that the median nerve (top arrow) is compressed and adherent to the flexor tendons (bottom arrow), which are surrounded by a thickened wall.

trophy with non-steroidal anti-inflammatory drugs and physical therapy for 10 months. No biopsy had been taken despite a previous history of tuberculosis, which had been evident on chest radiographs. The initial radiographs had shown slight soft tissue swelling on the palmar aspect of the wrist. Thickening around the flexor tendons was seen on a CT scan, and advanced-stage osteopenia was found on radiographs taken 4 months later.



Figure 2. Case 2. The right hand, at the time of admission, with progressive destruction of the carpus and the metacarpal bones and osteolytic lesions in the radius and ulna.



Figure 3. Case 2. Stabilization of bone destruction was evident at the 10-month follow-up. The whole carpus is definitely smaller.

The physical examination showed slight swelling of the dorsoradial extensor tendon sheaths of the right wrist; the skin was warm and red in this area. Wrist flexion, extension, and thumb opposition were severely limited with complete loss of ulnar deviation. The radiographs revealed extensive destruction of the metacarpal bones, carpus and forearm (Figure 2). Laboratory data showed a positive rheumatoid factor (RF), and increases in ESR, leukocyte count, C-reactive protein level and antinuclear factor (ANF). The tuberculin skin test and Ziehl-Neelson stain were negative. The positive rheumatoid factor and slightly elevated ANF were associated with inflammation of the wrist.

Tenosynovectomy of the extensor pollicis longus, extensor carpi radialis longus and extensor carpi radialis brevis muscles with removal of caseous material and partial wrist synovectomy were performed. The histopathological and bacteriological examinations showed progressive tuberculous synovitis. Wrist mobilization was started 4 days postoperatively. Drug susceptibility tests showing no antituberculous resistance were completed after the chemotherapy was started. Together with the internist, the patient was placed on a multidrug regimen, including pyrazinamide, ethambutol, rifampin, and isoniazid for 2 months, followed by a 3-drug combination of ethambutol, rifampin, and isoniazid for an additional 10 months, and by rifampin, and isoniazid for another 2 months. Regular follow-ups were done, and the pain and swelling subsided completely (Figure 3). 8 years postoperatively, the patient was clinically free of disease and was satisfied with the functional outcome.

## Discussion

Although the incidence of TB has steadily fallen in the industrialized countries worldwide since 1993 (World Health Organization 2000), cases of extrapulmonary involvement are common among immunosuppressed and elderly persons, and foreign-born persons from endemic areas (Watts and Lifeso 1996). Tuberculous tenosynovitis of the hand is uncommon and accounts for about 1% of cases with musculoskeletal TB (Pimm and Waugh 1957, Hodgson et al. 1972). It is usually caused by *M. tuberculosis*.

The disease is well described in reference books on hand surgery (Patel 1999). However, the literature abounds with cases of delayed or wrong diagnoses because of less awareness, particularly among young clinicians and surgeons who have not had contact with this disease, and the difficulty in rapidly establishing the diagnosis. Its insidious onset, initially nonspecific symptoms, radiographs, and laboratory data which may show a positive RF (Djavad et al. 1996) are frequently misdiagnosed as rheumatoid arthritis (Bush and Schneider 1984), reflex sympathetic dystrophy, tenosynovitis (Dahl 2001), Dupuytren's contracture (Jackson and King 1989), carpal tunnel syndrome (Regnard et al. 1996), soft tissue tumors, and chronic wounds (Cecidi et al. 1998). Intermittent pain relief is commonly achieved by inappropriate use of non-steroidal anti-inflammatory drugs. This disguises the clinical symptoms and delays the diagnosis. In elderly patients, the diagnosis of tuberculous tenosynovitis may be easily overlooked and confused with age-related or underlying illnesses (Rajagopalan and King 2001).

Any history of previous disease or of exposure to TB, and therapy-resistant tenosynovitis with a decrease in hand function are suggestive of extrapulmonary involvement (Serke et al. 1999). 50% of patients have abnormal chest radiographs (Serke et al. 1999) as we saw in our 2 patients. Surgical findings of chronic proliferative tenosynovitis, fluid with rice bodies or caseous masses are highly suggestive of TB (Pimm and Waugh 1957, Jaovisidha et al. 1996). Rice bodies may also occur in rheumatoid arthritis, but most of them are much smaller (Bush and Schneider 1984). Histological and bacteriological analyses are required to confirm the diagnosis (Legal and Pfeiffer 1971) and distinguish *M. tuberculosis* from atypical mycobacteria, such as *M. malmoense*, *M. marinum*, *M. kansasii*, *M. terrae*, *M. avium-intracellulare*, and the *M. fortuitum-chelonae* complex; most of these are preceded by trauma, and person-to-person transmission is less likely to occur (Woods and John 1987, Gabl et al. 1997). They have various clinical manifestations. *M. marinum* infection, also referred to as "swimming pool granuloma" or "fish tank granuloma", is associated with cutaneous tuberculosis which may develop after contact with water inhabited by fish or with contaminated

nonchlorinated fresh water, but tuberculous tenosynovitis may also occur. *M. marinum* has an optimal culture growth temperature of 31°–33 °C, but other mycobacteria grow at 37 °C (Woods and John 1987). This fact is quite important as many laboratories do not routinely incubate mycobacterial material at such low temperatures.

Improvements in nucleic acid amplification tests, such as DNA and PCR techniques, which are now commercially available, allow rapid detection of *M. tuberculosis* in cases with extrapulmonary involvement (American Thoracic Society 2000). At the time of diagnosing our 2 patients, these modern techniques of mycobacteriology were not available.

Tuberculous tenosynovitis, although relatively rare, should be considered in the differential diagnosis of recurrent tenosynovitis of the hand, especially when combined with risk factors, such as advancing age, impaired immunity, and previous residence in endemic areas. Early radical excision of the infected tissues combined with systematic antituberculous multidrug therapy gives good functional results and prevents a recurrence. Patients with extrapulmonary disease, in particular those with bone and joint TB, should receive a minimum of 12 months of therapy. Basically, the initial treatment consists of isoniazid, rifampin, and pyrazinamide given for 2–3 months followed by isoniazid and rifampin for 4 months in patients with very susceptible organisms. Ethambutol should be included in the initial therapy until the results of drug susceptibility studies are available, unless there is little likelihood of drug resistance (Serke et al. 1999). Interaction between the orthopedic surgeon and internist or pulmonologist is strongly recommended for individualized and successful treatment. Increased awareness and a high index of suspicion of extrapulmonary TB are mandatory in making an early diagnosis and thus preventing deformities and a reduction in function, as seen in our 2 cases.

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