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CASE REPORT

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Gouty flexor tenosynovitis of the hand mimicking atypical mycobacterial infection

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Abstract A 50-year-old Japanese fish dealer presented with painful and swollen fingers. Infectious flexor tenosynovitis with *Mycobacterium marinum* was suspected. Range of motion was restored after tenosynovectomy and after ofloxacin and clarithromycin were administered. Two years after the operation, the patient presented again with acute inflammation in the same fingers. Histopathological examination revealed gouty tenosynovitis. The preconception that mycobacterial infection occurs often in fish dealers caused us to miss the correct diagnosis of gouty tenosynovitis.

Key words Gout · Hand · Tenosynovitis · Tophus · Urate crystal

Introduction

Tenosynovitis of the flexor tendons of the hand arise from several conditions or situations such as overuse, trauma, infection, crystal deposits, tumor, rheumatoid arthritis, hemodialysis, and diabetes mellitus. Infectious causes include purulent bacteria, tuberculous and nontuberculous mycobacteria, and fungi. In particular, nontuberculous mycobacterial tenosynovitis in fish breeders is well known as fish-tank granuloma. On the other hand, gouty tenosynovitis is a rare entity, and lack of awareness of this condition can result in an incorrect diagnosis.

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Case report

A 50-year-old Japanese man visited our outpatient clinic because of swelling of his right little and ring fingers and the left little finger that had lasted for 1 month. He was a fish dealer and had repeated small lacerations of his fingers caused by fish fins and scales. Physical examination revealed spindle-shaped swelling and very slight tenderness in the proximal phalanx along the flexor tendon without redness and no local heat (Fig. 1a,b). The active range of motion (ROM) of the proximal interphalangeal joint (PIP) of the right ring finger was limited from –10° in extension to 50° in flexion and that of the metacarpophalageal joint (MP) was limited from –20° to 60°, so that the fingertip could not make contact with the palm. Plain radiographs showed no bone abnormalities or ectopic calcification in the fingers and wrist (Fig. 2). Laboratory data were within the normal range except for the erythrocyte sedimentation rate being 55 mm/h and C-reactive protein (CRP) 0.3 mg/dl. Rheumatoid factor was within the normal range. The uric acid level was not determined. The physician thought the patient might have infectious flexor tenosynovitis with *Mycobacterium marinum*, so tenosynovectomy was performed. A tuberculin PPD skin test was negative.

Intraoperative findings revealed white powdery deposits in the subcutaneous adipose layer and synovial sheath that had infiltrated both flexor tendons (Fig. 3). Deposits were removed and annular and cruciform pulleys were excised, leaving a part of A2 and A4 pulleys. The initial pathological finding was granuloma formation with multinucleated giant cells. Atypical mycobacterium infection could not be ruled out. Acid-fast staining, culture under a low temperature (30°C), and polymerase chain reaction (PCR) using a specific nucleotide for atypical mycobacterial species did not reveal any bacilli infection.

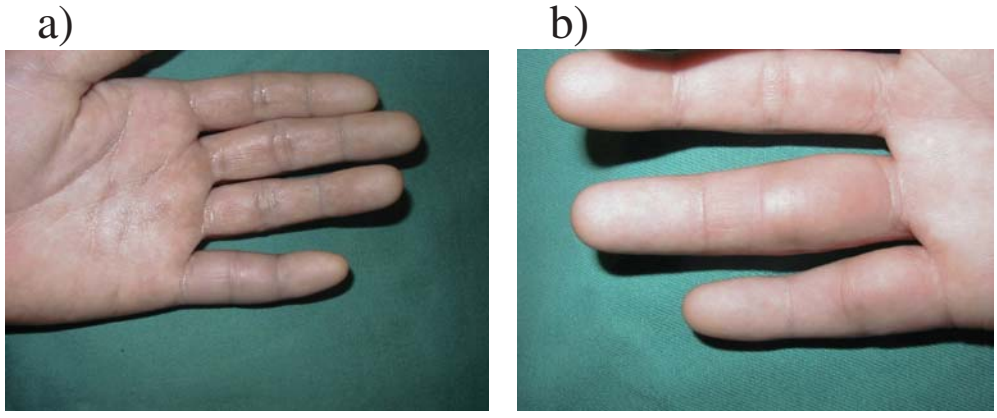

Fig. 1a,b. Appearance of hands at the first visit. Photos show swelling of the left little finger (**a**), and right ring and little fingers (**b**). There is no redness that would suggest acute inflammatory change

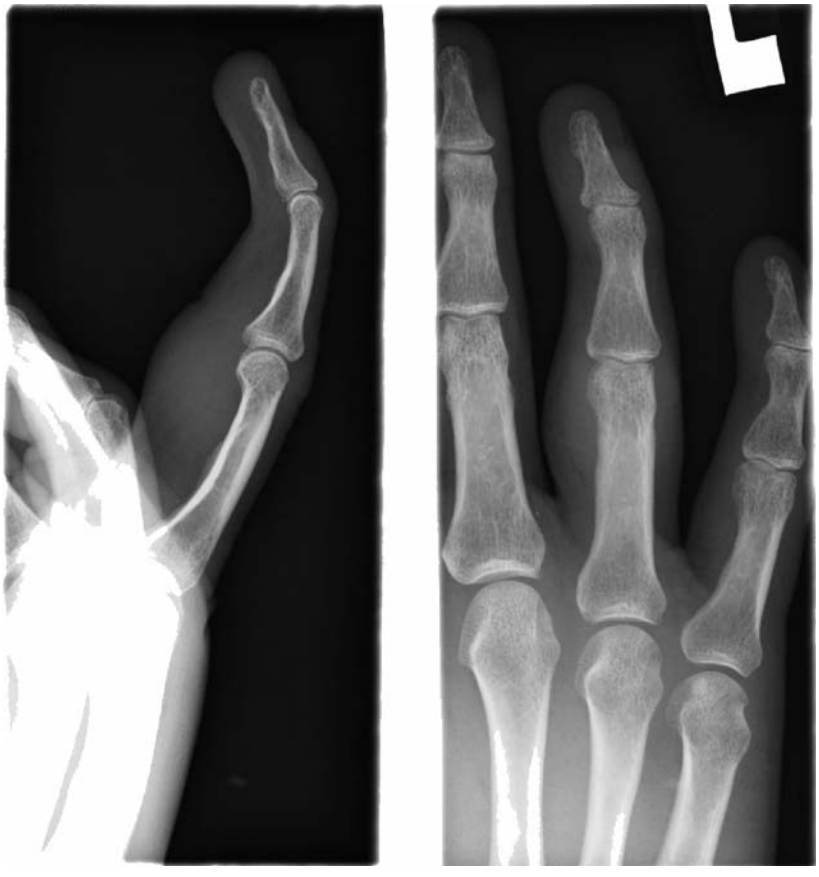

Fig. 2. Plain X-ray film shows no ectopic calcification in the finger but evidence of soft tissue swelling

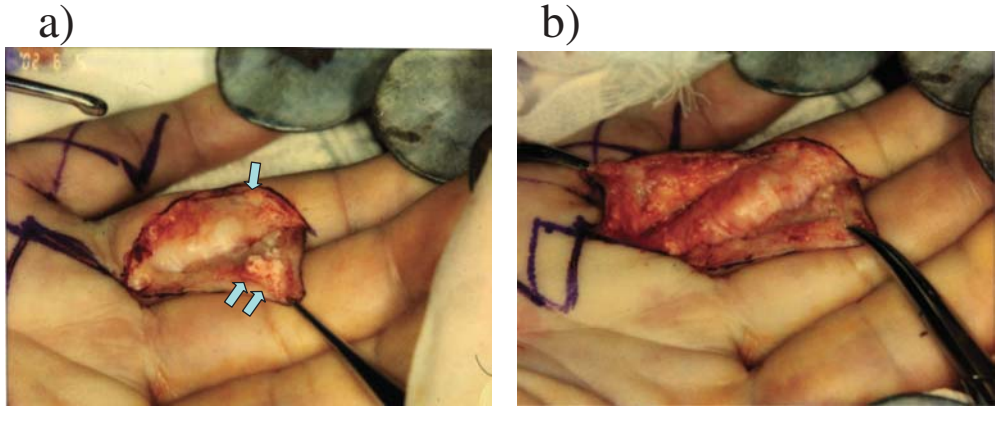

Fig. 3. During the operation, the incision disclosed white chalky deposits (*arrows*) in the subcutaneous layer (**a**), and diffuse swelling of the tenosynovial sheath at the level of middle and proximal phalanges (**b**)

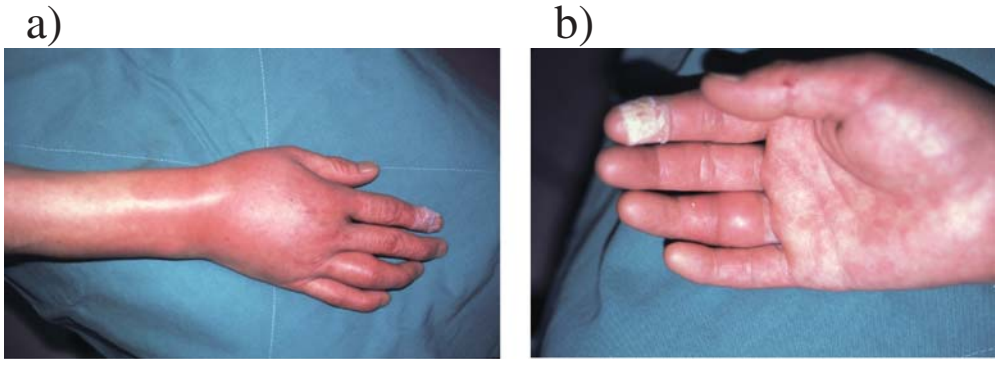

Fig. 4a,b. Appearance of right hand at the second visit 2 years after surgery. Diffuse swelling with severe redness especially in the dorsal side of the hand (**a**), and volar aspect of proximal phalanx of the ring finger (**b**) are evident

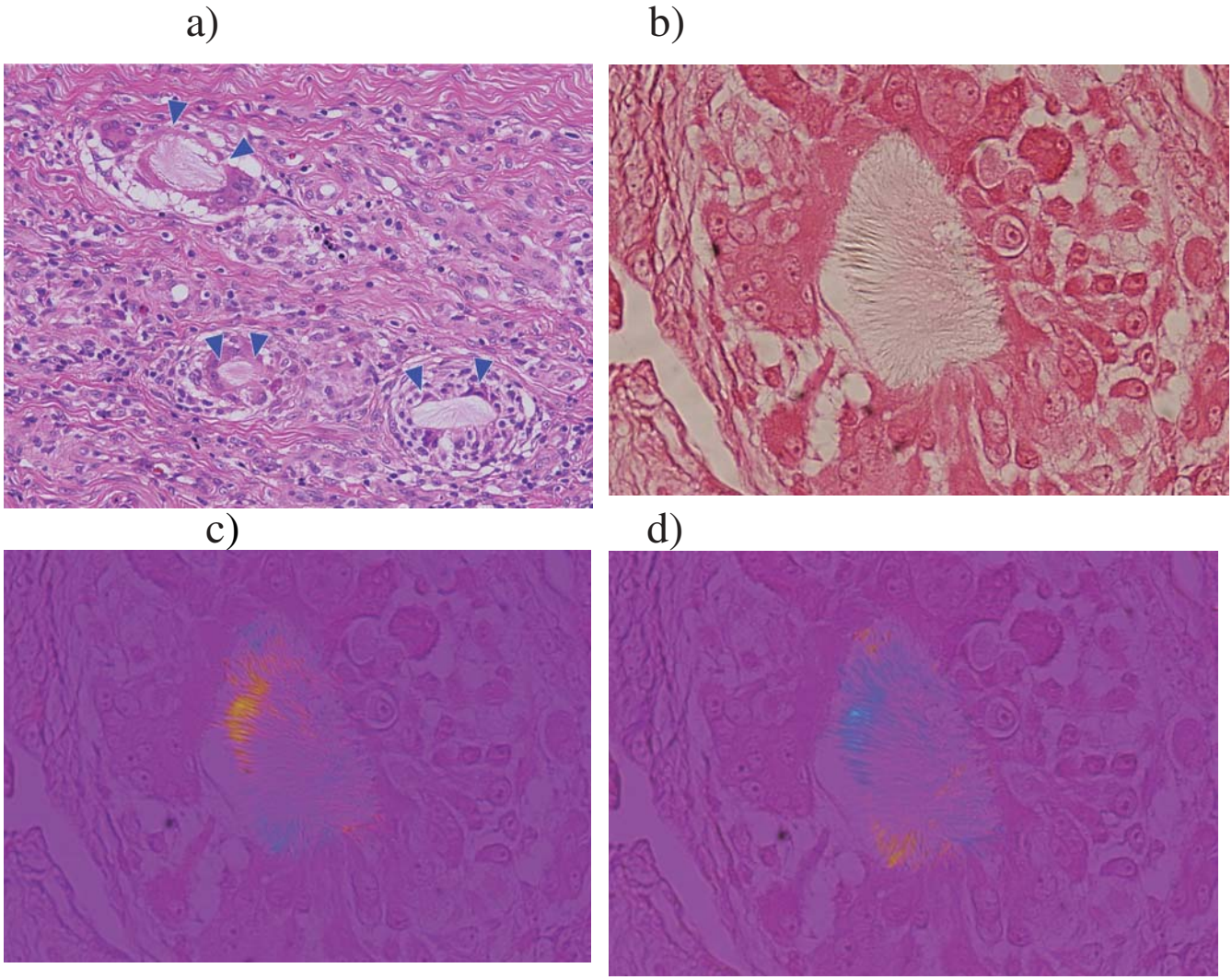

Fig. 5. a Histopathologically, the tenosynovium (formalin-embedded section) shows a foreign body reaction including multinucleated giant cells and epithelioid cells. Foreign bodies consist of slightly eosinophilic material with a thin slit-like pattern (*arrowheads*). These findings are compatible with the tophus that was fixed by formalin, and needle-shaped urate crystals were eluted (H&E stain). There were no infectious agents, such as fungi, bacteria, or acid-fast bacilli, by special stains such as PAS, Gram, and Ziehl-Neelsen. **b** A plethora of needle-shaped crystals is observed at the center of foreign body granuloma. The periphery of the crystals shows a light tan color (Eosin stain). **c,d** Polarized microscopic view. The crystals show negative birefringence under polarized light. The crystals parallel to the line of slow vibration appear as brilliant yellow, whereas those at right angles to the line of slow vibration are brilliant blue (**c**). When the sensitive color plate (red-plate compensator) is rotated through 90°, the color of each crystal, yellow and blue, changes to the opposite of that which they were initially (**d**). These features are pathognomonic for urate crystals[1]

After the operation, the patient was administered antibiotics (ofloxacin 300 mg and clarithromycin 400 mg per day) for 2 weeks. Swelling and pain resolved day by day, and active ROM of the PIP and MP improved to the right angle in flexion. The patient voluntarily discontinued medical examinations.

Two years after the initial medical examination, the patient complained of severe pain, redness, and swelling of his right ring finger. He had not experienced any pain, swelling, redness, or loss of ROM during the intervening 2-year period. Severe swelling, redness, and local heat were present on the dorsal site of the hand and ring finger (Fig. 4a,b). Limitation of both active and passive ROM of the PIP had markedly recurred. The patient declared that he had a history of gout going back 5 years. His uric acid level was 8.8 mg/dl (normal range: 2.3–7.3 mg/dl), CRP 4.8 mg/dl (normal upper limit: 0.3 mg/dl), and white blood cell count 15 400/μl (normal range: 4000–8000/μl). An aspirated specimen did not suggest appropriate crystals of monosodium urate monohydrate infiltration. Physical examination revealed no evidence of tophus at any other location of the body. However, re-examination of the former surgical material by a different pathologist disclosed tophi in the resected tissue (Fig. 5a). For the detection of urate crystals, formalin-fixed paraffin embedded tissue was cut to a thickness of 5 μm, quickly extended on a water bath, and scooped onto a glass slide. After drying, the slide was placed in xylene and absolute ethanol, respectively, for 5 min. To avoid solving out of the uric acid, the slide was stained by eosin in 70% ethanol, dehydrated by 80%, 90%, and 100% ethanol and xylene, and was mounted by coverglass using regular mounting medium. The slide was evaluated under a polarizing microscope. Several areas had radial aggregates of abundant needle-like crystals in the center of granuloma, and the crystals showed negative birefringence. These findings were consistent with tophi from urate crystals (Fig. 5b–d).[1]

The patient was administered a nonsteroidal anti-inflammatory drug (loxoprofen sodium, 180 mg, three times a day) while the inflammation was sustained. Two weeks later, pain and redness had faded. He is still under treatment with allopurinol. Plication of flexor tendons is considered because of ROM insufficiency.

Discussion

Flexor tenosynovitis is caused by infection, tumor, rheumatoid arthritis, gout, and other conditions. Atypical mycobacterium infection occurs frequently in workers who handle fish, and this is well known as fish-tank granuloma among hand surgeons. It is generally a superficial infection, but sometimes may invade into a deeper layer. Zenone et al.[2] pointed out in their review that ten different species of nontuberculous mycobacteria have been found to cause tenosynovitis, with *M. marinum* the most frequent cause. The diagnosis of atypical mycobacterial infection should be made by culture under suitable conditions or PCR with species-specific probes; however, Laing et al.[3] reported a case in which both cultures with swab and aspirate were negative. Histopathological studies show several variations in inflammation with or without granuloma and with or without caseous necrosis,[4] and in which giant cells were of both the Langhans and foreign body type. Harth et al.[5] suggested in a review that delay in diagnosis and initial inappropriate treatment with intra-articular steroids were frequent. At the time of our patient's first visit, there was no evidence of acute inflammation. These preconceptions misled us to make the diagnosis of an atypical mycobacterial infection.

The diagnosis of gouty tenosynovitis is not easy, because its symptoms are not specific and can present as an infection, tendon rupture, nerve compression, or digital stiffness.[6] To make the diagnosis more difficult, the serum uric acid level is often within the normal limit on the occasion of a gouty attack.[7]

Radiological examinations are not very useful. Yu et al.[8] evaluated magnetic resonance (MR) images from nine gouty patients and described the MR appearance of tophaceous gout as nonspecific. Therefore, they did not recommend the use of MR imaging in the initial diagnostic evaluation of patients with gout.

Walker and Dawn[9] stated that repeated aspiration of the suspicious site or open biopsy is necessary to establish the correct diagnosis, but careful inspection is necessary because formalin elutes urate crystals, and specimens should be fixed in ethanol for their detection. However, as we successfully demonstrated, the characteristic birefringence of the urate crystals can be visualized under a polarization microscope equipped with a red-plate compensator, even in the formalin-fixed paraffin-embedded tissue.

Granulomatous infiltration with multinucleated giant cells raises the possibility of infection with mycobacteria, certain fungi, *Brucella* species, or other infectious agents. More attention should be paid to the fact that these infections could histopathologically mimic gouty tenosynovitis. Some reports[10–14] point this out and all emphasize awareness of the possibility of gouty tenosynovitis. Abrahamsson[10] reported a case that was initially treated as infectious tenosynovitis but at reoperation gouty deposits were found penetrating the flexor tendon sheath. Townshend and Pai[13] noted that gout mimicking infectious tenosynovitis may be more common than is currently recognized.

The reason is obscure as to why the symptoms on the second visit were more severe than those at the first presentation, but it is conceivable that urate crystals shed from microtophi could lead to an acute gouty attack. Surgical intervention can precipitate gout. Calderon and Chung,[15] in fact, reported a case of carpal tunnel release triggering a gouty attack 2 weeks after surgery. In our case, however, the period between synovectomy and the emergence of hand inflammation was more than 2 years, so it could not be supposed that the intervention precipitated the attack directly.

Treatment of gouty tenosynovitis is almost always surgical because exploration and open biopsy are necessary to establish the correct diagnosis. Also, gouty tenosynovitis of

the hand may present with a variety of signs and symptoms including compression neuropathy,[16] infection, soft tissue mass, joint stiffness,[17] or signs of tendon rupture.[18,19] These conditions require drainage, tenosynovectomy, neurolysis, mass resection, joint mobilization, and tendon repair or tendon transfer. Immediate postoperative physical therapy is important.

We presented a case of gouty tenosynovitis that was originally misdiagnosed as atypical mycobacterial infection. When infectious tenosynovitis is suspected, gouty tenosynovitis should be included in the differential diagnosis.

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