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CASE REPORT

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Subcutaneous tendon rupture of extensor tendons on bilateral wrists associated with calcium pyrophosphate dihydrate crystal deposition disease

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Abstract Calcium pyrophosphate dihydrate (CPPD) crystal deposition disease is a well-recognized inflammatory joint disorder. Extensor tendon rupture associated with CPPD deposition has rarely been described. We report herein the case of a 58-year-old woman who underwent reconstruction for subcutaneous extensor tendon ruptures of the extensor tendons for the ring and little fingers on both wrists associated with CPPD deposition. The significance of this case is the occurrence at an earlier age compared to previous papers and the appearance on bilateral wrists.

Key words Arthropathy · Calcium pyrophosphate dihydrate (CPPD) · Tendon rupture · Tophaceous deposition · Wrist

Introduction

Calcium pyrophosphate dihydrate (CPPD) crystal deposition disease is a well-recognized inflammatory joint disorder. The pathophysiology involves the accumulation of CPPD crystals and can be confused with degenerative osteoarthritis.

Treatment for this disease usually involves nonsteroidal anti-inflammatory drugs (NSAIDs). The literature contains few descriptions of CPPD deposition disease in multiple joint replacement surgeries. In addition, no reports have described subcutaneous extensor tendon ruptures on bilateral wrists in patients with CPPD deposition disease. We report herein the case of a patient who underwent not only total joint arthroplasties of bilateral knees and the right hip, but also reconstruction for subcutaneous rupture of the extensor tendons for the ring and little fingers on bilateral wrists.

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Case report

A 58-year-old woman presented to our hospital in February 1998 complaining of an inability to actively extend the metacarpophalangeal joint of the left ring and little fingers. She had experienced sudden and painless loss of extension ability for these fingers 2 weeks earlier. No history of trauma, rheumatoid arthritis (RA), Colles' fracture, or overuse of the hand was elicited. Furthermore, no other associated joint problems were identified at that time. Medical history included intracranial bleeding in April 1996, diagnosed on brain computed tomography (CT). Physical examination revealed a healthy-appearing woman in no acute distress. The left wrist was swollen and slightly tender without redness or local heat. General joint laxity was observed, with thumb to the arm, elbow hyperextension and knee recurvatum. Plain radiography of the left wrist showed calcification in the triangular fibrocartilage complex (TFCC).

In May 1998, surgery was performed under general anesthesia. The extensor tendons for the ring and little finger had ruptured completely at the wrist without fibrous connective tissue strands. The dorsal capsule was worn out, through which thick synovial tissue was come out. Crystal deposition was noted in the synovium and the TFCC associated with bony erosions at the radio-ulnar joint. After the removal of proliferated synovium, a Sauve–Kapandji procedure[1] (distal radio-ulnar arthrodesis with distal ulnar pseudoarthrosis) was performed. Ruptured tendons could not be sutured end-to-end, and so were sutured side-by-side to the extensor tendon of the middle finger. Wrist and fingers were immobilized in a splint for 3 weeks. The postoperative course was uneventful, and the patient was able to extend the ring and little fingers satisfactory as of the time of writing. Histological examination revealed chronic synovitis with foreign body giant cell reaction to crystals.

On compensated polarized light microscopy, crystals exhibited weakly positive birefringence. CPPD deposition disease was diagnosed and treatment with NSAIDs was initiated.

In September 1998, the patient presented to our clinic with new pain in bilateral knees. Plain radiography of the left knee showed calcium deposition on the meniscus and bone destruction in the lateral compartment (Fig. 1). Laboratory studies revealed a C-reactive protein (CRP) level of 0.20 and rheumatoid factor (RF) level of 3.1. All other laboratory studies were within normal range. Right and left total knee arthroplasties (TKAs) were performed in September 1998 and September 1999. Histological examination revealed chronic synovitis with CPPD crystals. In May 2003, the patient again presented to our clinic with new pain in the right hip. Plain radiography of the right hip showed osteoarthritis with joint space narrowing and periarticular cyst formation. Right total hip arthroplasties (THAs) were performed in October and December 2004, at 63 years old. Histological examinations again revealed chronic synovitis with CPPD crystals.

The patient presented to our hospital once more in August 2006, describing an inability to actively extend the metacarpophalangeal joint of the right ring and little fingers. Laboratory studies revealed a CRP level of 0.20 and RF level of 3.0, and a matrix metalloproteinase (MMP)-3 level of <20 ng/ml. The left wrist was swollen and slightly tender without redness or local heat. Plain radiography of the right wrist showed destruction and subluxation of the radio-ulnar joint with calcification in the radiocarpal joint. Degenerative changes were seen in the metacarpophalangeal joint of the thumb and trapezioscaphoid joint (Fig. 2). Surgery was performed under general anesthesia. The extensor tendons of the ring and little fingers on the right wrist had completely ruptured without fibrous connective tissue strands. The dorsal capsule was worn out, exposing thick synovial tissue. Crystal deposits were noted in the synovium and TFCC associated with bony erosion at the radio-ulnar joint. In addition, tenodesis of a slip of the extensor carpi ulnaris tendon to the distal ulna was performed to avoid a symptomatic unstable proximal ulnar stump. After the proliferated synovium was excised, a Sauve–Kapandji procedure was performed. The radial and ulnar articular surfaces of the distal radio-ulnar joint were decorticated using osteotomes. After removing a 10-mm segment of ulna with surrounding periosteum, permanent fixation of the distal radio-ulnar joint was obtained using an absorbable screw. The ruptured tendons could not be sutured end-to-end, and so were sutured side-by-side to the extensor tendon of the middle finger. Wrist and fingers were immobilized in a splint for 3 weeks. Histological examinations demonstrated chronic synovitis with CPPD crystals surrounded by macrophages and giant cells (Fig. 3). The postoperative course was uneventful, and the patient was able to extend the ring and little fingers satisfactorily as of the time of writing.

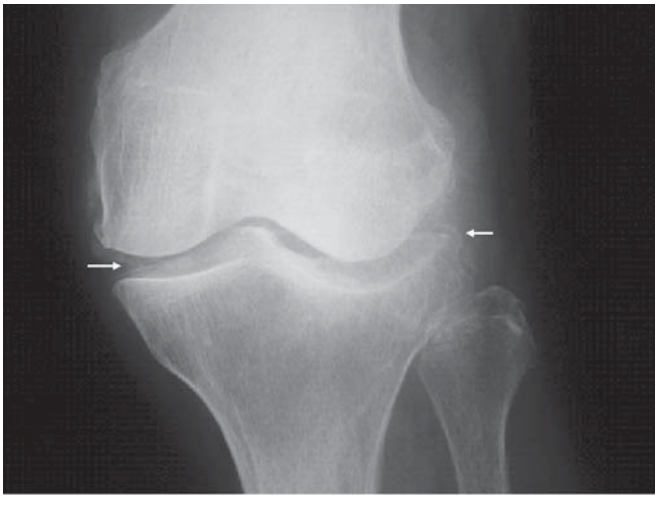

Fig. 1. Plain radiography of the left knee shows calcium deposition on the meniscus (*arrows*) and bone destruction of the lateral compartment. Involvement of the lateral compartment of the knee, which is rarely seen in ordinary degenerative osteoarthritis, represents a distinctive feature of calcium pyrophosphate dehydrate (CPPD) arthropathy

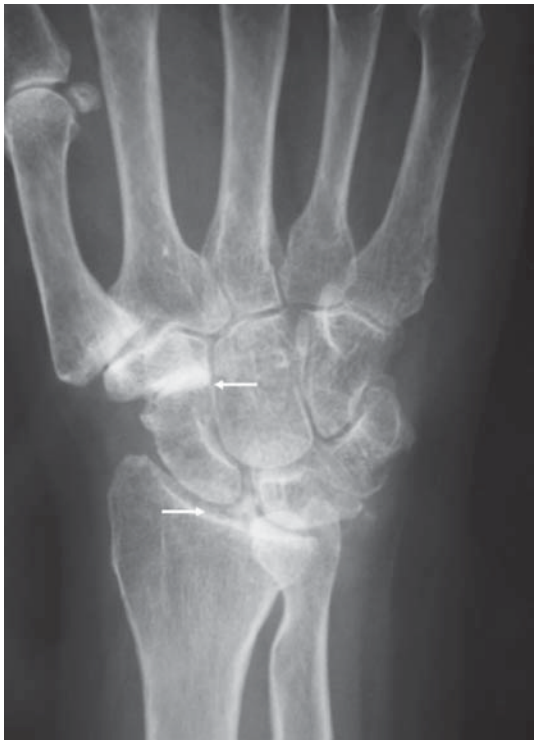

Fig. 2. Preoperative radiography of the right wrist shows calcification in the radio-carpal joint (*arrows*), prominent at the ulnar head. Degenerative changes were seen at the metacarpophalangeal joint of the thumb and trapezioscaphoid joint. Involvement of the trapezioscaphoid joint, rarely seen in ordinary degenerative osteoarthritis, is a distinctive feature of CPPD arthropathy

Discussion

McCarty et al. identified CPPD crystals in joint fluid and established the concept of pseudogout, representing acute inflammatory arthritis.[2] McCarty then reported various patterns of clinical manifestation for joint diseases associated

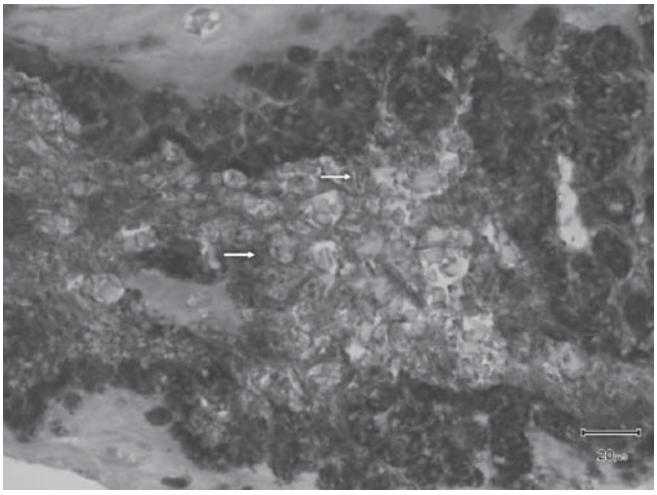

Fig. 3. Deposition of CPPD crystals in wrist synovium removed at surgery. Depositions (*arrows*) were surrounded by macrophage and giant cell reaction (H&E, ×25)

with CPPD crystal deposition.[3] He reported a mean age of 72 years for patients at the time of diagnosis for CPPD crystal deposition disease. Resnick et al. reported that although alterations superficially resembled osteoarthritis, CPPD crystal deposition disease was frequently more severe and progressive with extensive fragmentation of bone, causing intra-articular osseous bodies, and also that the disease occurred in unusual locations, such as the radiocarpal compartment of the wrist.[4] Various disorders related to CPPD crystal deposition in the upper extremity have been reported, including destructive wrist arthroplasty[5] and carpal instability.[6] However, subcutaneous rupture of extensor tendons associated with CPPD crystal deposition is rare, with only eight reported cases, all in women.[7–12] Compared to the age distribution for previously reported cases (mean age, 77.7 years), our patient is the youngest and the only case with bilateral tendon ruptures. In addition, marked local bone and joint destruction occurred at both knees and right hip, resulting in bilateral TKAs and right THA. Our patient displayed general joint laxity, which might have caused carpal instability and osteoarthritis. Tendon ruptures of the extensor tendons in bilateral wrists might have occurred at an earlier age in association with CPPD crystal deposition disease and general joint laxity.

Although deposition of CPPD crystals in the tendon sheath associated with tenosynovitis has been described, the exact mechanisms leading to rupture remain unclear. Tendon ruptures may be related to not only crystal-induced tenosynovitis, but also distal radio-ulnar joint instability, which causes attrition of extensor tendons in the wrist. Surgical treatment should thus be a combination of tendon repair with synovectomy and Sauve–Kapandji procedure to preserve pronation and supination.

Chronic crystal-associated arthropathies can lead to local bone destruction.[3] CPPD crystals initiate cartilage destruction by simulating mitogenesis of synovial lining cells, in addition to synthesis and secretion of proteases, prostanoids, and cytokines that have been implicated in cartilage matrix degeneration. Currently available drugs for treating CPPD crystal-induced arthritis, including NSAIDs and glucocorticoids, are predominantly directed towards the control of pain and inflammation in joints, but do little to limit joint destruction. The excessive inflammatory responses in joints are caused by proinflammatory mediators from residential cells stimulated by crystals. Inhibiting signaling pathways in these cells thus seems logical. Interaction of CPPD crystals with monocytes/macrophages leads to induction of a number of pro-inflammatory cytokines, such as interleukin (IL)-1, -6 and -8, and tissue necrosis factor alpha (TNFα).[13–15] In addition, stimulation of these cells with crystals reportedly activates Src-family kinases, which then mediate activation of MAPK and NF-κB pathways, eventually leading to nitric oxide (NO) generation and expression of IL-18 and MMP-3.[15–17] Interrupting these signaling pathways may have therapeutic potential for inhibiting crystal-induced joint inflammation. Biological agents such as anti-TNFα and IL-1 antibodies, which can inhibit MAPK and NF-κB activation, have recently been used to ameliorate progression of bone destruction in RA.[18–22] These agents can thus offer more effective treatment for crystal-induced bone and joint destruction. In addition, Hirose et al.[23] reported that increased cartilage intermediate-layer protein and ankylosis protein expression participate in excess accumulation of extracellular inorganic pyrophosphate and promote CPPD crystal formation in articular cartilage, indicating that these proteins may offer potential targets for CPPD crystal-induced arthritis treatment. Further studies of the microcrystal-induced signaling pathways leading to inflammation and joint destruction and the molecular mechanisms of articular microcrystal deposition will yield new insights into the development of more effective treatments for crystal-induced arthritis.

Our findings in this case show that CPPD crystal-induced synovitis in the wrist joint can lead to rupture of the extensor tendons, which should be recognized as a complication of CPPD crystal deposition disease even in middle-aged patients. The Darrach procedure[24] was considered as an option, but the Sauve–Kapandji procedure was thought to offer the best solution for stabilizing the distal radio-ulnar joint with ulnocarpal support.

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