Upper limb pyrophosphate tenosynovitis outside the carpal tunnel

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SUMMARY Three cases of calcium pyrophosphate dihydrate (CPPD) crystal deposits in tendon sheaths outside the carpal tunnel are reported. Crystals were shown by x ray diffraction analysis in one case and by compensated light microscopy in the other two. Surgical excision of the tendon synovial sheath had to be done in two cases (one case with CPPD crystal deposits).

Extra-articular deposition of calcium pyrophosphate dihydrate (CPPD) crystals has been described in rare cases, especially in the ligamentum flavum and in various kinds of tendons: triceps, flexor digitorum, and Achilles tendon. It has also been reported in bursitis, in subcutaneous tissue, and even in the dura mater. Presumed similar deposits have been radiologically reported in patients otherwise known for CPPD crystal deposition disease—for example, in transverse ligament of the second cervical vertebra and in flexor digitorum. The association of carpal tunnel syndrome with articular chondrocalcinosis has been classically reported; deposits of CPPD crystals have been found in the volar carpal ligament as well as in the tendon sheaths.

We report the finding of CPPD crystals in the synovial fluid from tendon sheaths outside the carpal tunnel.

Case report

CASE 1
A 72 year old woman had been operated on for a bilateral carpal tunnel syndrome 10 years ago. She had been suffering from pain in the left knee for about five years and from pain in both shoulders for three years. In addition, she had had painful swelling of the ventral part of the fifth left finger for about 10 months. Clinically, the tendon sheath was diffusely swollen and tender and motion of the finger was restricted (Fig. 1). The left knee was swollen and tender and the shoulders were limited and tender. Radiographs showed chondrocalcinosis of both wrists, both knees, and a destructive arthropathy of the left shoulder. Degenerative

Fig. 1 Case 1. Diffuse swelling of the tendon sheath of the fifth finger.
changes of the first carpometacarpal joints were also found in the left hand.

Examination of the synovial fluid of the left knee showed $0.2 \times 10^9$ leucocytes/l with rare intracellular positive birefringent crystals. Red S alizarin staining was negative. Synovial fluid was removed from the tenosynovial cavity of the finger (25 January 1988); it was gelatinous and contained rare white deposits 2-3 mm in diameter. x Ray powder diffraction analyses of this material showed CPPD crystals in triclinic form. Tenosynovitis recurred, and a synovectomy was performed in April 1988. Histological examination was performed on slides stained by haematoxylin and eosin and by von Kossa stain for calcium phosphate. It showed a non-specific scar remodelling of the synovium with gelatinous deposits and only an isolated small area of CPPD crystal deposits. Recovery was uneventful.

CASE 2
In February 1988 a 77 year old man developed nodular thickening of the palmar surface of his left wrist. He was also suffering from pain in the first carpometacarpal joints bilaterally and from chronic pain in the left knee with some acute typical attacks of pseudogout. Examination of the left wrist showed a distended synovial sheath of the flexor carpi ulnaris 0.8 cm in diameter.

There were no signs of carpal tunnel syndrome. x Ray examination showed chondrocalcinosis of both knees but no chondrocalcinosis of the wrists.

Aspiration of the synovial fluid from the tenosynovial cavity produced a clear fluid containing $1.3 \times 10^9$ leucocytes/l (98% mononuclear) with plenty of intracellular positive rod-like birefringent crystals. A topical corticosteroid agent was injected with a good result.

CASE 3
A 53 year old woman had suffered from tenosynovitis of the extensor tendon sheaths of the fingers in 1965. A tenosynovectomy was performed, but there was no histological examination. In 1986 the symptoms occurred again.

Clinical examination was normal except for a swollen tendon sheath of the extensor tendons of the right wrist and for a tenderness of the first carpometacarpal joint. Examination of 0.5 ml of synovial fluid from the tendon synovial cavity showed $0.9 \times 10^9$ leucocytes/l (98% mononuclear) with intracellular and extracellular positive rod-like birefringent crystals.

An x ray examination of the hand and knees failed to show signs of chondrocalcinosis. A tenosynovectomy was performed in November 1986. The synovial fluid contained crystals. Histological examination was performed on stained (haematoxylin and eosin and von Kossa stain) and unstained slides, which were examined by polarised light. It showed a non-specific synovial hyperplasia with scar remodelling but without CPPD crystal deposits. Examination of some fragments by transmitted electron microscopy and by x ray electron diffraction did not show CPPD crystals.

Discussion
The common feature of these three cases is a hand tenosynovitis whose exudated synovial fluid contained CPPD crystals which were disclosed by x ray diffraction in one case and by the presence of rod-like positive birefringent crystals in the others. In only one case were CPPD crystal deposits found in the neighbouring synovium and this patient had presented with radiological signs of chondrocalcinosis in both knee and wrist joints. In the other two cases chondrocalcinosis was shown by x ray examination in only one of them.

These cases show that CPPD crystal deposits in the hand in extra-articular locations induce not only a carpal tunnel syndrome but also tenosynovitis even in the absence of radiological signs of chondrocalcinosis elsewhere in the joints. Synovial fluid examination seems to be more simple and sensitive than a synovial biopsy, particularly as histological changes are not equally spread. It is uncertain whether the primary CPPD crystal deposits involve the tenosynovial membrane or the tendon itself.

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References
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