

CASE REPORT

Akimasa Kobayashi · Toshiro Futami · Isao Tadano
Mamoru Fujita

Spontaneous rupture of extensor tendons at the wrist in a patient with mixed connective tissue disease

Received: August 24, 2001 / Accepted: January 25, 2002

Abstract A 54-year-old woman who had been treated for mixed connective tissue disease for 4 years developed spontaneous rupture of extensor tendons in the wrist. The patient was surgically treated by tendon reconstruction. Histopathological examination of the synovial membrane showed lymphocytic inflammatory cellular infiltration around small blood vessels. The tendon ruptures in this case were most likely caused by synovial membrane proliferation in the wrist and mechanical stress generated by the subluxated distal ulna.

Key words Extensor tendon · Mixed connective tissue disease (MCTD) · Spontaneous rupture · Tendon reconstruction · Wrist joint

Introduction

Sharp et al.¹ first described mixed connective tissue disease (MCTD) as a syndrome showing clinical symptoms of systemic lupus erythematosus (SLE), polymyositis, and systemic sclerosis, and characterized by the presence of antibodies to extractable nuclear antigen. However, among patients with MCTD, some demonstrate clinical findings of polyarthritis with accompanying radiological bony changes^{2,3} which are similar to those found in rheumatoid arthritis (RA). The most commonly affected joint is the wrist.⁴ We report a rare case of spontaneous extensor tendon rupture in a patient treated for MCTD, in which radiographic findings showed bony changes in the radiocarpal joint and dorsal subluxation of the distal ulna. The causes of tendon rupture are discussed.

Case report

A 54-year-old right-handed woman was referred to our department because of an inability to use her right hand. In 1995, the patient developed swelling and pain in both wrists and Raynaud's signs, together with arthralgia in both shoulders and pain in both arms. She visited the Department of Collagen Disease in our hospital in 1996, and MCTD was diagnosed from clinical symptoms such as arthralgia, muscle weakness, Raynaud's phenomenon, and sclerodactyly, and a positive test for antiribonucleoprotein (RNP) antibodies at a titer of 216. Since then, she had been treated with prednisolone for 4 years, at a total dose of approximately 12 g. In October 1999, she had difficulties in fully extending her right ring and little fingers, but had not sought treatment. In January 2000, she was incapable of extending the right middle finger when opening the lid of a bottle. Because of the inconvenience with using her right hand, she was referred to our department.

On examination, active extension of the metacarpophalangeal (MP) joints was -45° in the middle finger, -80° in the ring finger, and -90° in the little finger. On radiographic examination, a posteroanterior view in the right wrist showed sclerotic change in the radiocarpal joint, marked spicule formation on the radius at the ulnar side, and 3 mm of ulna plus variant. A lateral view showed erosion on the dorsal side of the carpal bones and dorsal subluxation of the distal ulna (Fig. 1). Blood tests showed an erythrocyte sedimentation rate of 60 mm, a C-reactive protein level of 1.5 mg/dl, a positive rheumatoid factor, and positive anti-RNP antibodies. Other blood tests were within the normal ranges.

Surgery was conducted in April 2000. Intraoperative findings revealed complete rupture of the third, fourth and fifth extensor digitorum communis (EDC) and extensor digiti minimi (EDM), and also proliferation of synovial membrane (Fig. 2). Surgery was performed in the order synovectomy, distal ulna resection, and tendon reconstruction, which consisted of tendon transfer of the extensor indicis proprius to the EDM, and a tendon graft of the

A. Kobayashi (✉) · T. Futami · I. Tadano · M. Fujita
Department of Orthopaedic Surgery, Kitasato University East
Hospital, 2-1-1 Asamizodai, Sagami-hara 228-8520, Japan
Tel. +81-42-748-9111; Fax +81-42-748-5120
e-mail: aki-ko@kitasato-u.ac.jp

Fig. 1a,b. Radiographic findings of the right wrist. **a** The posteroanterior view shows mild narrowing of the radiocarpal joint, marked spicule formation on the radius at the ulnar side, and 3 mm of ulna plus variant. **b** The lateral view shows erosion on the dorsal side of the carpal bones and dorsal subluxation of the distal ulna

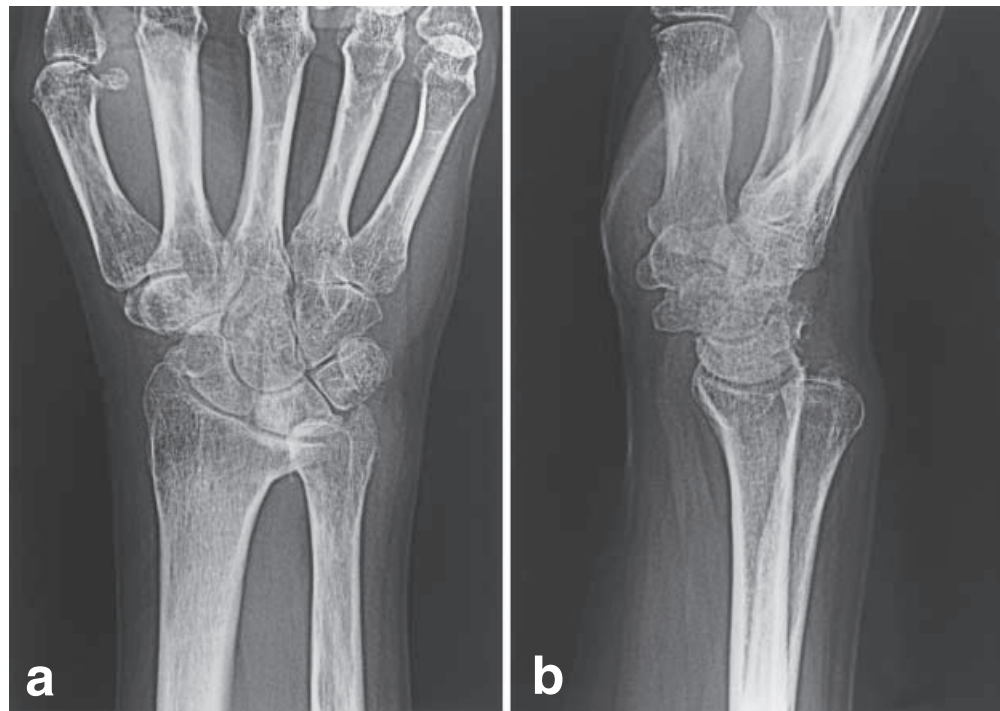


Fig. 2. Intraoperative findings. A proliferated synovial membrane and complete rupture of the extensor tendons can be seen

palmaris longus for the EDC. The hand was immobilized in a plaster splint for 3 weeks after surgery. Histopathological examination of the synovial membrane showed inflammatory cellular infiltration, composed mainly of lymphocytes, around the small blood vessels (Fig. 3).

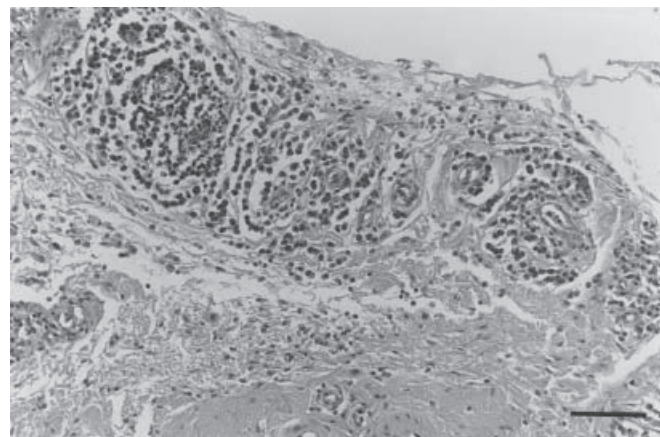


Fig. 3. Histopathological findings of the synovial membrane. Inflammatory cellular infiltration, composed mainly of lymphocytes, is seen around small blood vessels. Bar 100 μ m (HE staining)

One year after surgery, active extension of the third, fourth and fifth MP joint was -15° , and the patient's grip strength was 110 mmHg (130 mmHg with the unaffected hand). There was no inconvenience in the activities of daily living.

Discussion

Spontaneous rupture of extensor tendons in the wrist is rare and occurs mainly in RA,⁵ but also in SLE,⁶ osteoarthritis of the distal radioulnar joint,⁷ Kienbock disease, and distal radius fracture. The major causes of extensor tendon rup-

ture in rheumatoid wrists have been understood to be tendon invasion by the proliferated synovium and/or bony changes due to chronic synovitis. These factors lead to attrition of the surrounding extensor tendon.

Bony changes in the wrist have been documented in MCTD.⁸ However, as far as we are aware, there are no reported cases of MCTD with spontaneous rupture of an extensor tendon in the wrist. In the present case, radiographic findings of the wrist showed changes in the radiocarpal joint and dorsal subluxation of the distal ulna. Intraoperative examination revealed proliferation of synovial membrane in the wrist. These observations suggested that tendon ruptures in this case, which resembled those seen in rheumatoid wrists, were caused by the proliferated synovial membrane and/or mechanical stress generated by the subluxated distal ulna. However, histopathological examination of the synovial membrane showed inflammatory cellular infiltration composed mainly of lymphocytes around small blood vessels, which indicates progressive systemic sclerosis, and therefore differs from RA. This patient had arthralgia and positive rheumatoid factor, so there is the possibility of complications from RA. However, from the histopathological findings of the synovial membrane, we considered that the bony change in the affected wrist joint was due to MCTD.

This case required surgical treatment, which included distal ulna resection, together with adequate wrist synovectomy and tendon reconstruction. After these procedures, good hand function was regained.

References

1. Sharp GC, Irwin WS, Tan EM, Gould RG. Mixed connective tissue disease: an apparently distinct rheumatic disease syndrome associated with specific antibody to an extractable nuclear antigen (ENA). *Am J Med* 1972;52:148-59.
2. Udoff EJ, Genant HK, Kozin F, Ginsberg M. Mixed connective tissue disease: the spectrum of radiographic manifestations. *Radiology* 1977;124:613-8.
3. O'Connell DJ, Bennett RM. Mixed connective tissue disease - clinical and radiological aspects of 20 cases. *Br J Radiol* 1977;50:620-5.
4. Bennett RM, O'Connell DJ. The arthritis of mixed connective tissue disease. *Ann Rheum Dis* 1978;37:397-403.
5. Ryu J, Yuho Y, Nakamura K, Tanaka K, Toriyama S. Review of extensor tendon ruptures in the rheumatoid hand (in Japanese). *J Jpn Soc Surg Hand* 1987;4:244-9.
6. Khan MA, Ballou SP. Tendon rupture in systemic lupus erythematosus. *J Rheum* 1981;8:308-10.
7. Carr AJ, Burge PD. Rupture of extensor tendons due to osteoarthritis of the distal radio ulnar joint. *J Hand Surg* 1992;17-B: 694-6.
8. Halla JT, Hardin JG. Clinical features of the arthritis of mixed connective tissue disease. *Arthritis Rheum* 1978;21:497-503.