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CASE REPORT

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Flexor tenosynovitis of the hands as an initial manifestation of systemic lupus erythematosus

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Abstract We describe a patient who presented with flexion contractures of the bilateral fingers due to tenosynovitis of the flexor digitorum tendons as an initial manifestation of systemic lupus erythematosus (SLE). A 17-year-old woman had abrupt onset of diffuse swelling and flexion contractures in the bilateral fingers, accompanied by polyarthritis and cervical lymphadenopathy. Magnetic resonance imaging (MRI) showed flexor tenosynovitis of the hands. A diagnosis of SLE was made by immunological and hematological tests, and treatment with oral corticosteroids resulted in a rapid and complete disappearance of the flexion contractures.

Key words Flexion contracture · Flexor tenosynovitis · Systemic lupus erythematosus

Introduction

The musculoskeletal system is commonly involved in systemic lupus erythematosus (SLE). Polyarthritis, one of the major manifestations of SLE, shows mild arthralgia and synovitis and mostly absent erosive changes on radiographs, in contrast to those of rheumatoid arthritis (RA).[1,2] Joint deformities without bone destruction sometimes develop in the long course of the illness.[3] We describe a patient who, as an initial symptom and the chief complaint of SLE, manifested an abrupt onset of flexion contractures of the digits due to flexor tenosynovitis of the hands, which completely disappeared after treatment with corticosteroids. Although flexor digitorum tendons are frequently affected in RA, no detailed records of flexor tenosynovitis as an initial manifestation in SLE have been reported.

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Case report

A 17-year-old Japanese woman presented with swelling, tenderness, and limitation of extension in the left fingers in July 1998. The symptoms appeared abruptly, and a similar symptom developed in the right fingers 2 days later. She eventually developed arthralgia of the shoulders and elbows, backache, and general fatigue. She was seen by a orthopedician in August 1998, but treatment with nonsteroidal anti-inflammatory drugs and physiotherapy did not relieve her symptoms. She was referred to our hospital for further evaluation of the articular symptoms.

On admission, she complained of polyarthralgia of metacarpophalangeal (MP) joints, elbows, shoulders, knees, and ankles, with mild and short-duration (less than 30 min) morning stiffness. Examination showed flexion contractures of the bilateral fingers at the proximal interphalangeal (PIP) joints that was not correctable passively (Fig. 1). The second, third, and fourth fingers were severely affected and flexed nearly 90°, but the thumbs and little fingers were less affected. The proximal portions of the fingers were diffusely swollen, and MP joints were mildly swollen and tender. There were tender spots in both palms close to the MP joints, presumably along the flexor digitorum tendons. Raynaud's phenomenon, sclerodactylia, or nail lesion were not observed. Mild tenderness was noted in the right elbow, bilateral shoulders, and knees. A multiple lymphadenopathy was observed in the neck, and there was a diffuse erythematous swelling in the right pretibial region. No abnormal signs were found in the head, chest, and abdomen.

Laboratory investigations showed lymphocytepenia with 940/mm^3, an erythrocyte sedimentation rate of 59 mm/h, and C-reactive protein of 0.67 mg/dl (normal <0.3). Urinalysis was normal. Antinuclear antibody (ANA) was 1280× (homogenous and speckled type), anti-ds DNA

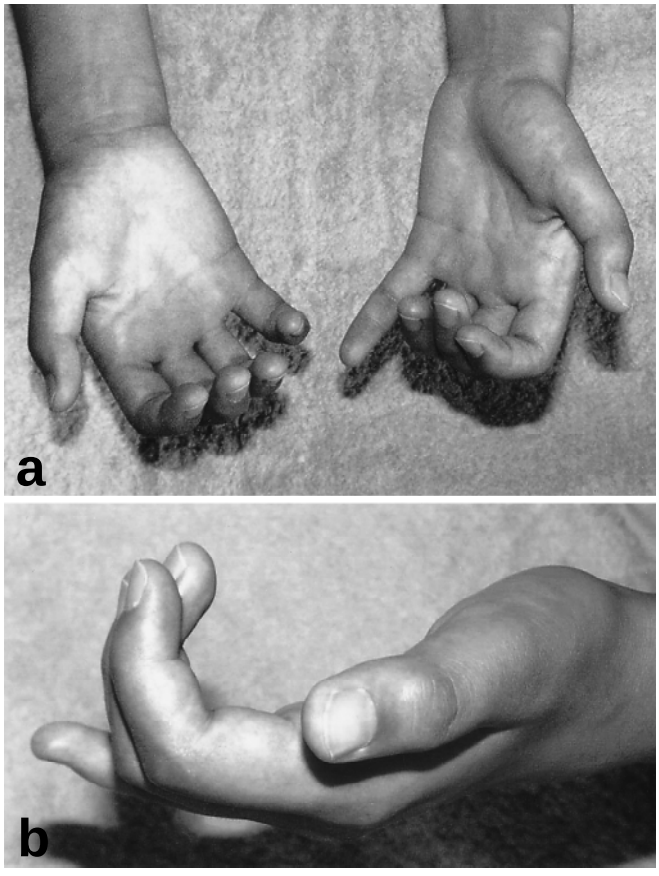

Fig. 1. **a** Photograph of the hands showing flexion contracture of the digits at PIP joints. **b** Lateral view of the left hand

antibody (ab) by ELISA was 674 IU/L (normal <12), and anti-SS-A ab by Ouchterlony immunodiffusion (OI) was 64×, but anti-Sm ab (ELISA), anti-RNP ab (ELISA), and anti-SS-B ab (OI) were negative. Rheumatoid factor (RF) as assayed by laser nepherometry, the latex test, and ELISA was negative. Severe hypocomplementemia with CH50 was 7 U/L (normal 30–55), C3 was 57 mg/dl (normal 80–170), and C4 was 5 mg/dl (15–40). Radiographs of the hands, wrists, elbows, shoulders, knees, and ankles were normal. Bone scintigraphy showed no significant accumulation of tracers in joints. MRI of the left hand showed high signal intensity areas around the flexor digitorum tendons within the proximal portion of the palm in T2-weighted images. These lesions were skipped along the tendons and showed strong enhancement by gadolinium contrast (Fig. 2). These findings indicated tenosynovitis (and peritendinitis) of the flexor digitorum tendons.

A diagnosis of SLE was made by polyarthritis, a high titer of ANA and anti-ds DNA ab, and lymphocytepenia, according to American College of Rheumatology (ACR) criteria.[4] Although no major organs, including the kidney, central nervous system, lungs, and heart, were involved, the patient had severe disability in her fingers with remarkable immunological abnormalities. Furthermore, no information about the course and management of acute onset flexion contractures in SLE could be obtained from the literature.

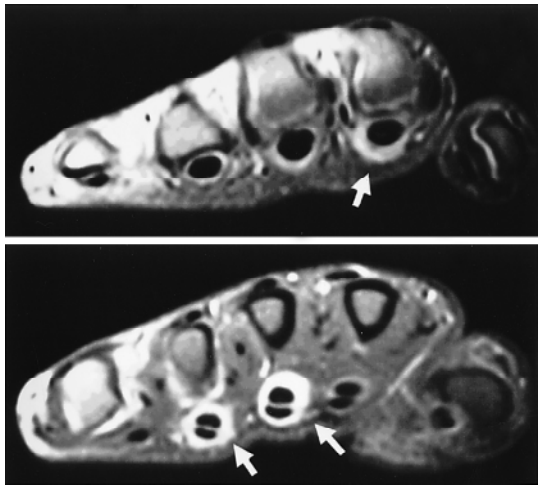

Fig. 2. MRI images of the left hand showing gadolinium-enhanced lesions around the flexor digitorum tendons. Note that the strongly enhanced tendons are different in the two scan levels

Therefore, we started to treat the patient with 40 mg/day predonisolone orally. Polyarthritis disappeared in 2 weeks, and flexion contractures and finger swelling gradually subsided and disappeared in 3 weeks. The results of laboratory tests were also improved by the treatment. Predonisolone was tapered, and the patient was discharged after 8 weeks' therapy. Since then she have been free of symptoms, and no contractures have been observed in digits. On her last visit in February 2000, 10 mg/day of predonisolone was prescribed, and no articular manifestations were observed. Laboratory tests showed anti-ds DNA ab of 29.9 IU/ml, CH50 of 25 U/L, C3 of 93 mg/dl, C4 of 10 mg/dl, and negative RFs.

Discussion

The present case manifested polyarthralgia and the sudden onset of flexion contractures of digits due to tenosynovitis, but typical features of SLE, such as butterfly rash, nephropathy, or serositis were absent. In this case, a differential diagnosis between SLE and RA came into question. We diagnosed the patient as having SLE according to the criteria of the ACR,[4] and in addition, hypocomplementemia was noted in the absence of rheumatoid vasculitis lesions. Furthermore, bone scintigraphy showed no focal accumulation in the hands despite serious symptoms. Recently, van de Wiele et al.[5] reported the usefulness of bone scintigraphy of the hands in differential diagnosis of early SLE and RA. They have shown that multiple foci of moderate to marked accumulation were observed in early RA, and that, on the other hand, normal findings or only a diffuse mild increase was observed in early SLE, suggesting mild synovitis in SLE.[5] These results supported our diagnosis. However, the possibility that SLE and RA overlapped could not be absolutely excluded in this case. Concurrence of SLE and RA, termed "rhupus," is not common.[6,7] This diagnosis has

been made on the basis of meeting both ARA criteria for RA[8] and ACR criteria for SLE, or by the existence of erosive and destructive arthritis in cases with SLE.[7,9,10] The present case failed to fulfill ACR criteria for RA,[11] and is only classified as probable RA by ARA criteria.[8] On this basis, the concurrence of RA is tentatively excluded in this patient, although careful follow-up observations are required. Another rheumatic disease that involves flexor digitorum tendons includes remitting seronegative symmetrical synovitis with pitting edema (RS_3PE syndrome).[12,13] This is a distinct form of polyarthritis, characterized by acute onset, symmetrical involvement, corticosteroid sensitivity, and a good course of illness. However, the absence of other characteristic features, such as late age at onset and pronounced pitting edema of the dorsa of both hands and feet, makes it less easy to diagnose the present case as being RS_3PE syndrome.

Uncommon articular manifestations of SLE include Jaccoud's arthropathy, contractures, tendon rupture, and carpal tunnel syndrome. Jaccoud's arthropathy is considered to develop in a long course of low-grade inflammation of joints and subsequent articular and paraarticular fibrosis.[14,15] Other deforming arthropathies of the hands, feet, and elbows can occur in patients with SLE, but most cases have a longer disease duration.[14–16] Reiley et al.[15] reported that flexion contractures in the digits had been observed by X-ray film in 26% of their SLE cases, but no detailed information about the patients was available. In contrast, three cases with carpal tunnel syndrome as a first manifestation of SLE have been reported,[17] in all of which the patients received local injections of corticosteroids and their symptoms were relieved in all cases. However, we were unable to find a case of SLE which presented with flexion contractures of the digits due to tenosynovitis as an initial manifestation, and which was successfully treated with corticosteroids. In RA patients, flexor tenosynovitis is one of the most common manifestations in hands. When it involves the proximal portion of the palm, snapping and locking of the digits occur, and tenosynovitis within the digits results in a loss of active flexion of those digits. In our case, MRI of the hand showed a skipped lesion of flexor tenosynovitis in the proximal portion, which induced flexion contracture of the digits. Although this may be a rare case, it should be noted that flexor tenosynovitis of the hands could be a distinct articular manifestation of SLE.

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