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Chronic bursitis and tenosynovitis revealing Whipple's disease

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Abstract

Joint complaints, most commonly intermittent arthritis, are the initial manifestation in about three-fourths of Whipple's disease cases. We herein report on two cases wherein Whipple's disease manifested itself as chronic bursitis and tenosynovitis at several sites.

A 42 year-old man had bilateral olecranon bursitis, a right patellar bursitis and an extensor tenosynovitis on the left wrist and a 54 year-old man had extensor tenosynovitis at both wrists and a bilateral tenosynovitis of the extensors at both ankle. Methotrexate in both patients and etanercept in one of them were not effective. Polymerase chain reaction testing revealed *Tropheryma whipplei* on feces, bursitis and articular fluid samples. Duodenal biopsy proved to be normal. Doxycycline and hydroxychloroquine were rapidly effective.

Chronic bursitis and tenosynovitis must be added to the list of rheumatologic manifestations that may evoke the diagnosis of Whipple disease.

Key words: Whipple disease, bursitis, tenosynovitis

Introduction

Whipple's disease is a chronic infectious systemic disease caused by the bacterium *Tropheryma whipplei*. Joint complaints are the initial disease manifestation in about three-fourths of cases. Clinically, the condition manifests itself by recurrence of arthritis in middle-aged men, less commonly by chronic arthritis, and, even more rarely, by spine or sacroiliac joint involvement [1]. We herein report on two cases wherein Whipple's disease manifested itself as chronic bursitis and tenosynovitis at several sites.

Cases

Case 1

A 42-year-old man was referred to our department for psoriatic arthritis refractory to methotrexate (MTX), with biological therapy being implemented. The symptoms had begun 18 months earlier with pain and swelling of the hands and ankles. The patient had been suffering from psoriasis for 10 years. Despite subcutaneous MTX (20mg/week), along with piroxicam, pain had spread to the shoulders and knees in symptomatic episodes lasting 2 to 3 days. Clinical examination revealed signs of bursitis involving both elbows and the right knee, along with extensor tenosynovitis affecting the left wrist. The erythrocyte sedimentation rate (ESR) was 8mm per hour, and the C-reactive protein (CRP) level was 28mg/L. The serology was negative for rheumatoid factor and for anti-CCP and antinuclear antibodies. The HLA phenotype was A3 A30 B44 B49 DR4 DR13. X-rays of the hands, forefeet, and pelvis were normal. Ultrasound imaging revealed an extensor tenosynovitis of the left wrist, along with a bilateral olecranon bursitis predominantly affecting the left side and a bilateral patellar bursitis predominantly affecting the right side. The fluid of the olecranon bursitis first

contained 8,100 leukocytes/ μ L, which increased to 17,000 leukocytes/ μ L, with 27% and 83% polynuclear neutrophils, respectively. Polymerase chain reaction (PCR) testing revealed *T whipplei* DNA in two feces samples, one saliva sample, and two bursitis samples. A duodenal biopsy proved to be normal upon histological examination, with a negative *T whipplei* PCR. The patient's management, consisting of doxycycline (200mg/day) and hydroxychloroquine (600 mg/day), was rapidly effective, with articular pain disappearing over 2 weeks, while the clinical examination, ESR, and CRP levels returned to normal within 3 months.

Case 2.

A 57-year-old man had suffered for 6 years from episodes of recurrent pain and articular swelling of the wrists and ankles lasting about 4 to 7 days. Clinical examination revealed signs of extensor tenosynovitis of the two wrists, flexor tenosynovitis of the third finger of the left hand and, at the ankles a bilateral dumb-bell tenosynovitis of the toe extensors. The articulations were not symptomatic. Ultrasound confirmed the presence of tenosynovitis, shown to be active on Doppler imaging, involving a synovitis of the left wrist, six metacarpophalangeal, tibiotalar and talonavicular joints, and three metatarsophalangeal joints, scarcely active upon Doppler imaging. The ESR was 7mm per hour, and the CRP was 15mg/L. The serology was negative for rheumatoid factor and anti-CCP and antinuclear antibodies. The HLA phenotype was A2 A26 B15 B35 DR4 DR15. X-rays of the joints, including the pelvis, were normal. Saliva, blood, and feces were all negative for *T. whipplei* by PCR. The patient was treated with hydroxychloroquine. Following a 3-month treatment, the articular pain episodes were still present, with similar signs of tenosynovitis upon clinical examination. Due to this, MTX was initiated. After temporary improvement, the articular pain episodes reoccurred. Eighteen months later, after negative PCR results for *T whipplei* in the saliva, blood, and feces, etanercept treatment was added to the pre-existing medication. Three months later, the improvement was quite marked, with less frequent episodes and fewer

involving severe pain, whereas the signs of poorly symptomatic tenosynovitis persisted upon clinical examination of the wrists and ankles. One year later, the patient suffered again from marked pain episodes, with a significant alteration of his general state of health, leading to hospitalization. Arthritis of the left knee was diagnosed, with a ruptured popliteal cyst into the calf. This time, *T. whipplei* PCR on a feces and a synovial fluid sample were positive. A duodenal biopsy proved to be normal on histological examination, with a negative *T. whipplei* PCR. The patient's treatment consisted of doxycycline (200mg/day) and hydroxychloroquine (600mg/day), which was markedly efficacious with the pain crises promptly disappearing, while clinical examination alterations and CRP levels returned to normal within 3 months.

Discussion

In these two cases, Whipple's disease was revealed by bursitis at several sites with tenosynovitis in one case and by tenosynovitis in several tendons in the other. The patients reported intermittent joint symptoms, though the only clinical sign observed upon initial clinical examination was the presence of chronic bursitis and tenosynovitis. These were manifestations of Whipple's disease, given that PCR for *T. whipplei* in bursitis fluid was positive in case 1 and most importantly, the clinical signs disappeared rapidly under antibiotic therapy, whereas they had persisted over months and even years following the implementation of antirheumatic therapy.

It must be noted that cases of bursitis and tenosynovitis are only rarely emphasized in rheumatologic series and updates pertaining to Whipple's disease [1-4]. In a series involving nine Whipple's disease cases, Olivieri *et al.* reported on two patients with olecranon bursitis, one patient with bilateral de Quervain's tenosynovitis lasting for one month, and five patients with episodes of swelling of the hands and feet, interpreted to be extensor tenosynovitis cases [5].

Chronic bursitis and tenosynovitis may increase the confusion with a rheumatic disease and the failure of conventional disease-modifying anti-rheumatic drugs often leads to the untimely implementation of biological therapy, as previously reported in the scientific literature [6]. PCR investigation of feces samples represents a very useful tool for detecting Whipple's disease but, as shown by our case 2 above, it is sometime necessary to perform multiple sampling at a temporal distance from any antibiotic therapy. Our cases had localized Whipple's disease, given that a duodenal biopsy proved to be normal and *in situ* PCR negative. The two patients cannot be considered healthy carriers because, aside from the feces samples, the PCR investigation was positive on synovial fluid in one case and on the fluid of bursitis in the other and, most importantly, a dramatic improvement was observed following initiation of antibiotic therapy. When a patient is seen outside of an arthritis episode and without any joint fluid available for analysis, sampling for PCR investigation can be performed on the fluid originating from the bursitis or tenosynovitis.

Conclusion

Chronic bursitis and tenosynovitis must be added to the list of rheumatologic manifestations that may evoke the diagnosis of Whipple's disease.

The authors state that they have no conflicts of interest with this article.

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