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CASE REPORT

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Achilles tendonitis in a patient with Behçet's disease

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Abstract A 56-year-old male patient had suffered from recurrent oral aphthae, acne-like rashes on the face, an erythema nodosum-like rash on the left lower leg, and severe heel pain on the left-hand side. Colonoscopy revealed six ulcerative lesions in the terminal ileum. Ultrasonography and magnetic resonance imaging showed an inflammatory lesion in the Achilles tendons, with greater inflammation on the left. Achilles tendonitis was considered to be a peripheral enthesopathy in this patient with Behçet's disease complicated by an ileal ulcer.

Key words Achilles tendonitis · Behçet's disease · Erythema nodosum · Intestinal ulcer · Ultrasonography

Introduction

Behçet's disease has been classified as a seronegative spondylarthropathy (SpA).[1] Achilles tendonitis has frequently been observed in cases of seronegative SpA as a symptom of peripheral enthesopathy.[1] However, there have been few reports of Achilles tendonitis as a complication of Behçet's disease.[2–4] Here, we describe the case of a patient who had suffered from recurrent oral aphtha and acne-like rash, with the recent development of an erythema nodosum-like rash, Achilles tendonitis, and the progression of intestinal ulceration. We also discuss the possibility that Achilles tendonitis can occur as a symptom of Behçet's disease.

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Case report

In February 2003, a 54-year-old male patient, who had been suffering from recurrent oral aphthae and acne-like rashes on the face, upper back, and chest, developed heel pain and achillodynia on the left-hand side, with a slight fever (37.5°C). Subsequently, heel pain also appeared on the right-hand side. The patient was treated by a home doctor with diclofenac sodium at a dose of 50 mg/day. In March 2003, the patient developed epigastralgia and painful nodal erythema in the left lower leg. Gastroduodenal endoscopy revealed a shallow ulcer in the anterior wall of the antrum. The patient was treated with sodium rabeprazole (20 mg/day) for the epigastralgia, and prednisolone (10 mg/day) for the persistent heel pain. In early April 2003, the patient was referred to, and admitted to, the Division of Rheumatology of Ohta Nishinouchi Hospital. A physical examination on admission showed extensive acne-like rashes on the face, neck, upper back, and chest. An erythema nodosum-like rash 2–3 cm in diameter with tenderness was observed at five sites on the left lower leg. Severe tenderness was observed at the left Achilles tendon. Because of the left heel pain, the patient could not touch his left heel to the ground when walking. Arthralgia was not observed in any joints. Other lesions, such as genital ulcer, anal ulcer, or uveitis, were not observed. Laboratory data on admission were as follows: white blood cells (WBC), 9100/μl; red blood cells (RBC), 420 × 10/μl; hemoglobin (Hb), 11.2 g/dl; platelets (PLT), 21.6 × 10/μl; erythrocyte sedimentation rate (ESR), 79 mm/h (normal value <10); C-reactive protein (CRP), 4.18 mg/dl (normal value <0.2); IgG, 1477 mg/dl (normal range 639–1349); IgA, 391 mg/dl (normal range 70–312); IgM, 106 mg/dl (normal range 56–352); hemolytic complement (CH50), 56.1 IU/ml (normal range 30–40); prothrombine time (PT), >100% (normal range 85–100);

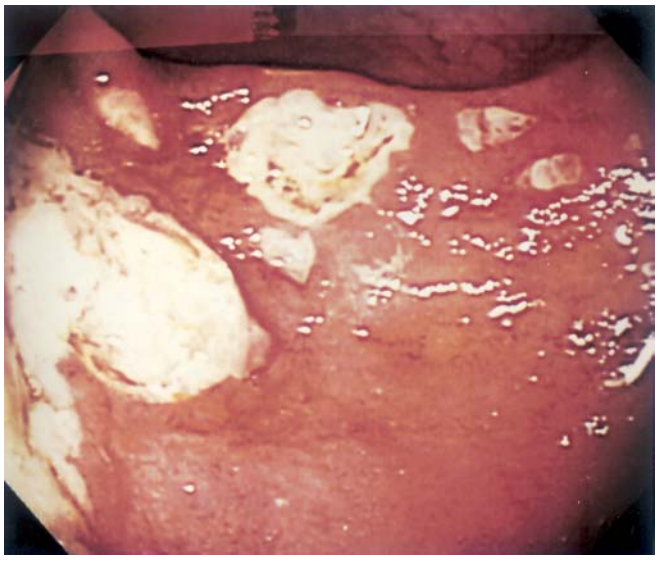

Fig. 1. Colonoscopic findings. Two large and four small ulcerative lesions with white coats sharply demarcated from the normal mucosa were observed in the terminal ileum

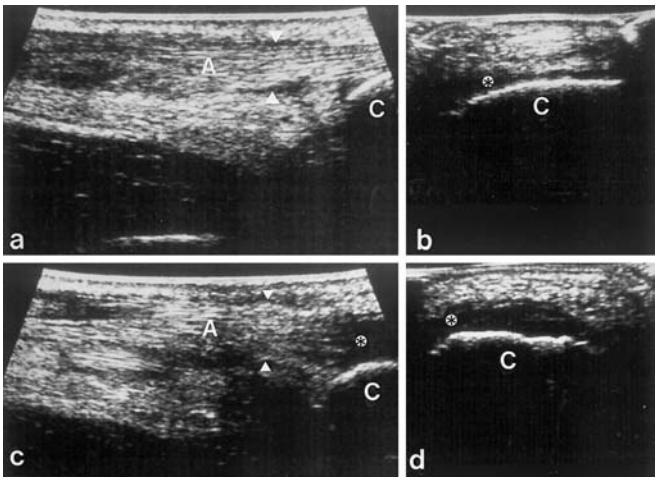

Fig. 2. Ultrasonography of Achilles tendons. **a** Longitudinal scan of the right Achilles tendon. A moderately echogenic fibrous slightly widened Achilles tendon (*A*) (▽△; width 9 mm) was observed. **b** Transverse scan of the right Achilles tendon. A low echogenic area (*; width 2.1 mm) was observed above the high echogenic line of calcanear cortex (*C*). **c** Longitudinal scan of the left Achilles tendon. A moderately echogenic fibrous slightly widened Achilles tendon (*A*) (▽△; width 10 mm) was observed. A wide low echogenic area (*) was observed between the Achilles tendon (*A*) and a high echogenic linear calcanear cortex (*C*). **d** Transverse scan of the left Achilles tendon. A wide low echogenic area (*; width 4.2 mm) was observed above the high echogenic line of calcanear cortex (*C*)

activated partial prothrombine time (APTT), 35.5 s (normal range 30–45); fibrinogen, 439 mg/dl (normal range 200–300). A pathergy test in the left forearm was positive on a 48-h evaluation. HLA typing results were A 11, A 31, B 54, and CW1. Rheumatoid factor and antinuclear antibody were not detected. A liver function test, renal function test, blood sugar, lipid analysis, electrolytes, and urinalysis were all normal. A chest simple radiography was normal. An abdominal ultrasound examination revealed a cystic lesion in the left kidney. Gastroduodenal endoscopy revealed chronic gastritis in the stomach. Colonoscopy revealed two large and four small ulcerative lesions with white coats demarcated sharply from the normal mucosa in the terminal ileum (Fig. 1). Pathohistology of the ulcerative tissue obtained by biopsy revealed nonspecific inflammation with an accumulation of a large number of small round cells. Two days after admission, an ultrasonographic examination of the Achilles tendons revealed bilateral widened Achilles tendons with a low echogenic area between the Achilles tendon and calcanear cortex, and with a greater change on the left-hand side than on the right-hand side (Fig. 2a,b). Similarly, magnetic resonance imaging (MRI) performed 24 days after admission revealed a high signal intensity in the Achilles tendons on a fat-suppressed and gadolinium-enhanced T1-weighted image (WI), with a greater change on the left-hand side than on the right-hand side (Fig. 3a,b). After a diagnosis of Behçet's disease, based on the International Study Group (ISG) criteria[5] (recurrent oral aphtha, erythema nodosum, and positive pathergy test), accompanied by an intestinal ulcer and Achilles tendonitis, colchicine (1 mg/day), loxoprofen sodium (180 mg/day), cilostazole (200 mg/day), salazosulfapyrizine (2000 mg/day), and minocycline hydrochloride (200 mg/day) were administered. The acne-like rashes and abdominal pain were rapidly relieved. The erythema nodosum-like rash and

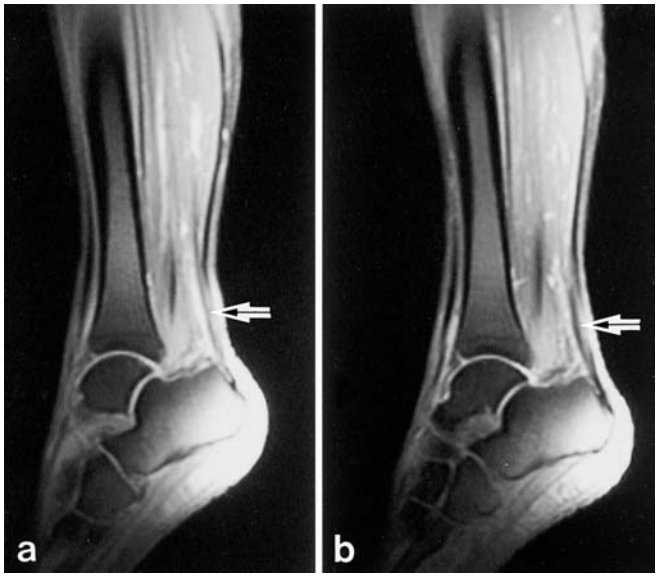

Fig. 3. T1-weighted, fat-suppressed, and gadolinium-enhanced magnetic resonance imaging scan of the Achilles tendons. **a** Sagittal scan of the right ankle. A high-signal-intensity area (←) was observed at the lower part of the Achilles tendon. **b** Sagittal scan of the left ankle. A high-signal-intensity area (←), with a greater intensity than on the right, was observed at the lower part of the Achilles tendon

achillodynia improved gradually, and acute-phase reactants decreased to within the normal range. The patient was discharged 24 days after admission. Colonoscopy performed in mid-June 2003 revealed no ulcer in the terminal ileum. At the last follow-up, in January 2004, the patient had no

skin lesions or achillodynia, but he exhibited occasional oral aphthae under the administration of colchicine and salazosulfapyrizine.

Discussion

These findings do not satisfy the standard Japanese diagnostic criteria for either the complete or the incomplete type of Behçet's disease (BD).[6] However, they satisfy the criteria of O'Duffy[7]) (recurrent oral ulcer, erythema nodosum, and colitis), the ISG[5] (aphtha, erythema nodosum, and a positive pathergy test), and Dilsen et al.[8] (recurrent oral ulcer, erythema nodosum, colitis, and a positive pathergy test). Achilles tendonitis is a symptom of peripheral enthesopathy, along with plantar fasciitis in seronegative SpA. The prevalence of Achilles tendonitis in patients with SpA ranges from 22%[1] to 58%.[9] The Achilles tendonitis of this patient was clearly demonstrated by ultrasonography and MRI on the left-hand side but not on the right-hand side. Behçet's disease might coexist with ankylosing spondylitis (AS).[10,11] In such cases, Achilles tendonitis may occur as a symptom of the coexisting AS. However, in the present patient, there were no findings of AS such as sacroiliitis, spondylitis, or the presence of HLA-B27. Here the intestinal ulcer in the terminal ileum was endoscopically typical of BD, i.e., a sharp edge demarcated from normal mucosa, which is quite different from the ulcers of Crohn's disease or intestinal tuberculosis. Then, histologically, there was no finding of granuloma formation in this ulcer, which is characteristic of Crohn's disease. Moreover, the pathergy test is usually negative in a patient with inflammatory bowel diseases. Therefore, this patient was diagnosed with BD associated with intestinal ulcer and accompanied by Achilles tendonitis. Intestinal ulcer in BD patients has reportedly been found more frequently in patients with incomplete type than complete type BD (52.8% vs. 29.6%).[12] The present case was classified as suspicious BD rather than incomplete BD, according to the standard Japanese criteria.[6] The various symptoms found in the present patient, such as aphtha, skin rashes, Achilles tendonitis, and intestinal ulcer, were improved sequentially by the treatment for BD, including colchicine, salazosulfapyrizine, and minocycline, without using corticosteroid. This suggests that these symptoms primarily originated from BD, and that some common mechanism induced Achilles tendonitis as well as the other symptoms. These findings support the hypothesis that BD belongs to a broader category of seronegative SpA.

In conclusion, Achilles tendonitis should be considered a possible symptom of Behçet's disease and other seronegative spondylarthropathies.

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References

1. Gerster JC. Plantar fasciitis and Achilles tendinitis among 150 cases of seronegative spondarthritis. Rheumatol Rehabil 1980;19: 218–22.
2. Caporn N, Higgs ER, Dieppe PA, Watt I. Arthritis in Behçet's syndrome. Br J Radiol 1983;56:87–91.
3. Paolaggi JB, Goutet MC, Stautz P, Siaud JR, Le Parc JM, Auquier L. Les enthesopathies des spondyloarthropathies inflammatoires. Frequence, description clinic, radiologique et anatomique. Etat actuel de la questions. A propos de 37 observations. Rev Rhum 1984;51:457–62.
4. Olivieri I, Cantini F, Napoli V, Braccini G, Padula A, Pasero G. Seronegative spondylarthropathy without spine involvement in Behçet's syndrome. Clin Rheumatol 1993;12:396–400.
5. International Study Group for Behçet's Disease. Criteria for diagnosis of Behçet's disease. Lancet 1990;335:1078–80.
6. Mizushima Y. Recent research into Behçet's disease in Japan. Int J Tiss React 1988;10:59–65.
7. O'Duffy JD. Suggested criteria for diagnosis of Behçet's disease [abstract]. J Rheumatol 1974;1:18.
8. Dilsen N, Konice M, Aral O. Our diagnostic criteria for Behçet's disease: an overview. Intern Cong Ser 1986;103:177–80.
9. Sambrook P, Mc Guigan L, Champion D, Edmonds J, Fleming A, Portek I. Clinical features and follow-up study of HLA-B27-positive patients presenting with peripheral arthritis. J Rheumatol 1985;12:526–8.
10. Borman P, Bodur H, Bostan EE, Barca N. The coexistense of Behçet's disease and ankylosing spondylitis. Rheumatol Int 2000; 19:195–8.
11. Kotevoglu N. Coexisting ankylosing spondylitis and Behçet's disease. Two cases with atypical presentation and course. Scand J Rheumatol 2003;32:184–5.
12. Baba S, Morioka S. Treatment of intestinal Behçet's disease. In: Inaba G, editor. Behçet's disease. Pathogenetic mechanism and clinical features. Tokyo: University of Tokyo Press; 1982. p. 559–70.