Reumatismo, 2019; 71 (4): 230-234

Subcalcaneal bursitis as the initial manifestation of rheumatoid arthritis: ultrasonographic observation of two cases

T. Suzuki, H. Shirai

Division of Allergy and Rheumatology, Japanese Red Cross Medical Center, Tokyo, Japan

SUMMARY

In early rheumatoid arthritis (RA), proliferative synovitis sometimes occurs earlier in the tenosynovium or bursal synovium than in the articular synovium. Here we report two patients who presented with subcalcaneal bursitis while progressing from undifferentiated arthritis with high-titer anti-CCP antibodies (ACPA) to a diagnosis of RA. They had initially presented with palindromic transient pain in the hands and the feet. They were strongly positive for ACPA and negative for rheumatoid factor (RF) at the onset of symptoms. A few years later, they developed persistent plantar heel pain and underwent musculoskeletal ultrasonography (MSUS). MSUS revealed subcalcaneal bursitis with synovial proliferation. At that time, they became positive for RF and they were clinically diagnosed and began receiving treatment for RA. They developed overt synovitis in their wrists and fingers several months later. To the best of our knowledge, this is the first report on MSUS-detection of subcalcaneal bursitis with synovial proliferation in patients in the very early phase of RA, although there have been many reports of forefoot bursitis. These cases suggest that MSUS scanning of the plantar surface of the heel may be useful for patients with plantar heel pain who are suspected of having a very early phase of RA, because proliferative synovitis can be detected as subcalcaneal bursitis.

Key words: Rheumatoid arthritis; subcalcaneal bursitis; ultrasound; plantar heel pain.

Reumatismo, 2019; 71 (4): 230-234

INTRODUCTION

Although plantar pain is often the earliest symptom of rheumatoid arthritis (RA), pain in the forefoot rather than in the hindfoot is usually reported (1). Here we report two patients who presented with subcalcaneal bursitis with synovial proliferation while progressing from undifferentiated arthritis with high-titer anti-CCP antibodies (ACPA) to a diagnosis of RA.

CASE REPORTS

Case 1

The patient in case 1 was a 43-year-old woman. Three years before her initial visit to our hospital, she underwent blood tests because she was concerned about a family history of RA despite the absence of symptoms. This test result showed a rheumatoid factor (RF) level of 2 U/mL (normal <15 U/mL) and an ACPA level of >100 U/mL (normal <4.5 U/mL).

She was subsequently referred to our hospital because of transient pain in her wrists and fingers. A physical examination did not show any sign of arthritis, although blood test results revealed that RF was 1 U/mL and ACPA was 546 U/mL. Because of the high ACPA titer, she had been followed regularly without treatment. While she had experienced episodic wrist pain, there were no swollen or tender joints during her visits.

Two years later, when she developed persistent pain and tenderness on the plantar surface of both of her heels, she underwent a musculoskeletal ultrasonography (MSUS) examination. The MSUS revealed peri-capsular hyperemia in the right second metacarpophalangeal (MCP) joint, which was the only sign of inflammation in her upper extremities, and bone

Corresponding author:
Takeshi Suzuki, Division of Allergy and Rheumatology, Japanese Red Cross Medical Center, Tokyo 150-8935, Japan
E-mail: takecsuzu@gmail.com

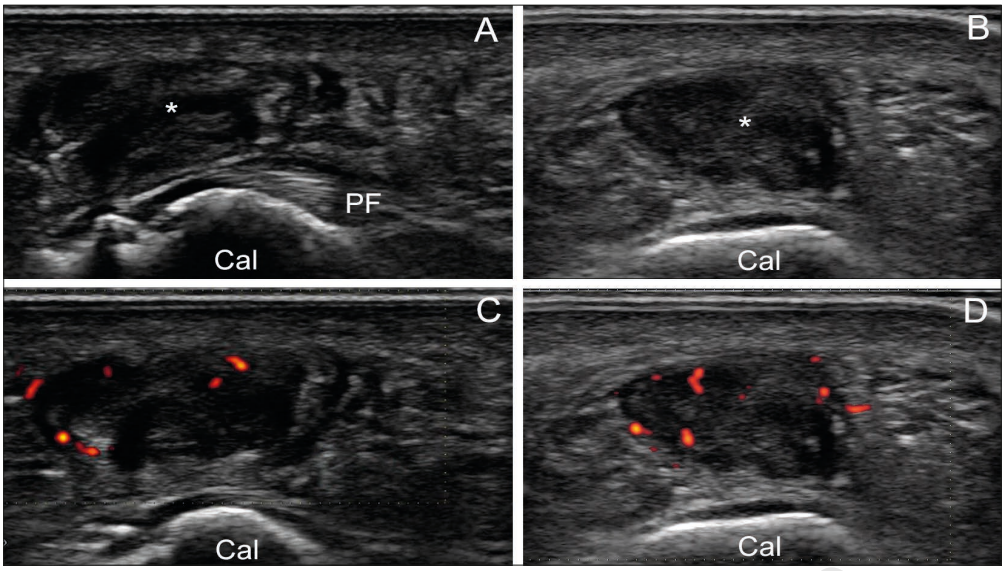

Figure 1 - Sonogram of subcalcaneal bursitis in the left plantar heel (Case 1). The long axis (A) and short axis (B) gray-scale sonograms show a well-demarcated predominantly hypoechoic and partially anechoic mass within the plantar fat pad. The long axis (C) and short axis (D) power Doppler sonograms show mild hyperemia at the margin and inside of the lesion. Cal, calcaneus; PF, plantar fascia.

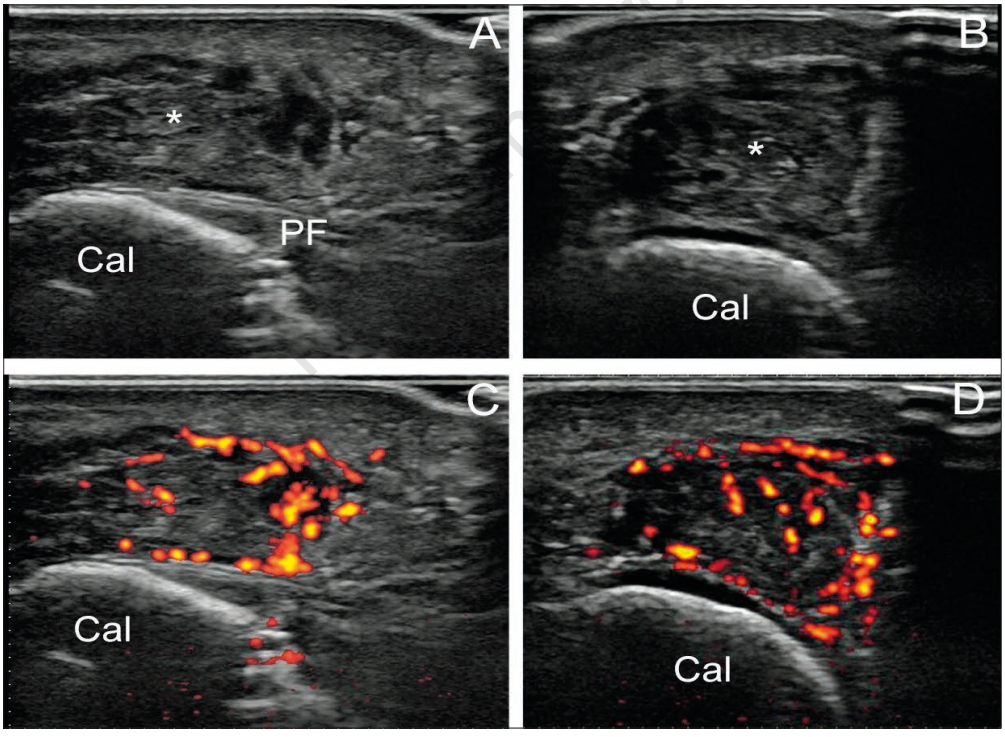

Figure 2 - Sonogram of the subcalcaneal bursitis in the left plantar heel (Case 2). The long axis (A) and short axis (B) gray-scale sonograms show a well-demarcated predominantly hypoechoic and partially anechoic mass within the plantar fat pad. The long axis (C) and short axis (D) power Doppler sonograms show moderate hyperemia at the margin and inside of the lesion. Cal, calcaneus; PF, plantar fascia.

erosion without active synovitis on the lateral aspect of the right fifth metatarsophalangeal (MTP) joint and mild synovitis in the right third MTP joint.
Additionally, predominantly hypoechoic lesions with mild hyperemia were detected within the plantar fat pad superficial to the calcaneal tuberosity in both of her heels (Figure 1). Since dynamic examination revealed that the mass was compressible by the ultrasound probe, a rheumatoid nodule was ruled out. Plantar fasciitis was also ruled out from the normal appearance of the plantar fascia. Because she did not regularly exercise, we concluded that the lesions were subcalcaneal bursitis presenting with a proliferation of the synovium that resulted from RA. She fulfilled the ACR/EULAR 2010 criteria for RA and methotrexate was started (2). At this time, RF had increased to 17 U/mL, which was a positive test result.
Six months later, she presented with swelling of the proximal interphalangeal (PIP) joints in her hands. Clinical remission was subsequently achieved following an increase in the methotrexate dose.

Case 2

The patient in case 2 was a 35-year-old woman. She had experienced episodic toe and plantar pain for over two and a half years at the time of her initial visit to our hospital. A physical examination did not reveal any sign of arthritis, although blood test results showed that RF was 8 U/mL and ACPA was 180 U/mL. Because of the high ACPA titer, she had been followed regularly without treatment. Ten months later, she developed episodic pain on the left plantar surface of her heel. Although swollen or tender joints were not observed at the time, her RF level had increased to 19 U/mL and ACPA had increased to 964 U/mL.
Six months later, she developed persistent finger pain and tenderness on the left plantar surface of her heel, and she underwent MSUS examination. The MSUS revealed peri-capsular hyperemia in the PIP joint of her bilateral second toe, which was the only sign of joint inflammation. Additionally, a predominantly hypoechoic lesion with moderate hyperemia was detected within the plantar fat pad superficial to the calcaneal tuberosity in her left heel (Figure 2). A rheumatoid nodule was ruled out because of its compressibility. Based on the lack of exercise habits, we concluded that the lesions were subcalcaneal bursitis presenting with synovium proliferation that resulted from RA. Although she did not fulfill the ACR/EULAR 2010 criteria for RA, methotrexate was started (2).
She fulfilled the criteria after one and a half months, when she developed synovitis in the MCP joints and the distal radioulnar joints and tenosynovitis of the extensor carpi ulnaris. Subsequently, clinical remission was achieved following the addition of subcutaneous golimumab.

DISCUSSION

The clinical courses of the two cases are very similar. They developed persistent plantar heel pain during the follow-up for high ACPA titers and palindromic arthralgia in their hands and feet. MSUS revealed subcalcaneal bursitis with synovial proliferation several months before they developed overt synovitis in their wrists and fingers.
ACPA is reported to be detectable in the serum before the onset of RA (3). We sometimes wonder whether we should start antirheumatic drugs for the patients with high ACPA titers who present with palindromic arthralgia/arthritis. If we can detect proliferative synovitis in such patients, initiation of therapy may be justified. MSUS seems to be the most feasible modality when searching for very mild or subclinical synovitis at various sites in the body for many reasons (4, 5).
The sites of the body that should be investigated for proliferative synovitis using MSUS in the patients with high ACPA titers and who are suspected of having a very early phase of RA have not been established yet. Based on the report comparing the early clinical features between RA patients with positive and negative ACPA results, seropositive patients had relatively more frequent involvement of MTP joints

compared with seronegative patients (6). Thus, patients who complain of foot pain should have their feet scanned. In the early phase of RA, proliferative synovitis sometimes or often occurs earlier in the tenosynovium or bursal synovium than in the articular synovium (1, 7-9). Therefore, in some cases, scanning the dorsal surface of the foot is not enough, and scanning the plantar surface of the foot is also required to detect proliferative synovitis in tenosynovium or bursa. Our observations suggest that proliferative synovitis can be also found as subcalcaneal bursitis in patients with plantar heel pain.

Although plantar heel pain is often associated with plantar fasciitis, which is frequently observed in seronegative spondyloarthritis, subcalcaneal bursitis is also another cause of plantar heel pain (10). The subcalcaneal bursa is thought to be an anatomical bursa with synovial lining that is located between the plantar fascia and the plantar fat pad, although information about this bursa is very limited and controversial (11). Magnetic resonance imaging and MSUS usually show fluid collection in the bursa in subcalcaneal bursitis that results from degenerative disorders (12). For RA patients, we searched and found only one case report describing subcalcaneal bursitis with synovial proliferation invading the calcaneus in long-standing active RA (13).

In the mid-2000s, however, Falsetti et al. published several reports describing a lesion called a *heel fat pad inflammatory edematous lesion* (14, 15). Although they initially reported it as a compressible, hypoechoic and poorly marginated area with peripheral vascularization (14), they subsequently reported that some lesions were well marginated and some vascular signals were also detectable inside some lesions (15). Since the lesions were located near the enthesis of the plantar fascia rather than throughout the entire thickness of the heel fat pad, we believe that the lesions called *heel fat pad inflammatory edematous lesions* are the same as the calcaneal bursitis that we described. We speculate that our more detailed observation enabled by advances in ultrasound technology may explain the different interpretations.

Falsetti et al. reported that the *heel fat pad inflammatory edematous lesions* were observed in 6.6% of 181 consecutive RA patients who were examined using MSUS, that the lesions were bilateral in half of the patients, and that the lesions were not associated with plantar fasciitis or enthesophyte in most cases (14). The lesions were also reported to be more frequent in patients with RA than in those with spondyloarthritis. Because the thick plantar horny layer and heel fat pad are becoming less of a barrier to ultrasound examination because of advances in the equipment, we consider that subcalcaneal bursitis is not an uncommon pathology that is detected by MSUS in patients with RA. However, because there are no reports about the frequency of subcalcaneal bursitis among the early RA patients, further investigations are needed to clarify the usefulness of MSUS scanning of the plantar hindfoot for the early diagnosis of RA.

CONCLUSIONS

To the best of our knowledge, this is the first report on ultrasound detection of subcalcaneal bursitis with synovial proliferation in patients in the very early phase of RA. MSUS-scanning of the plantar surface of the heel may be useful for patients with plantar heel pain who are suspected of having very early phase RA, because proliferative synovitis can be detected as subcalcaneal bursitis.

Conflict of interest

The authors declare no conflict of interest.

REFERENCES

1. Koski JM Ultrasound detection of plantar bursitis of the forefoot in patients with early rheumatoid arthritis. J Rheumatol. 1998; 25: 229-30.
2. Aletaha D, Neogi T, Silman AJ, et al. 2010 Rheumatoid arthritis classification criteria: an American College of Rheumatology/ European League Against Rheumatism collaborative initiative. Arthritis Rheum. 2010; 62: 2569-81.

3. Arend WP, Firestein GS. Pre-rheumatoid arthritis: predisposition and transition to clinical synovitis. Nat Rev Rheumatol. 2012; 8: 573-86.
4. Colebatch AN, Edwards CJ, Østergaard M, et al. EULAR recommendations for the use of imaging of the joints in the clinical management of rheumatoid arthritis. Ann Rheum Dis. 2013; 72: 804-14.
5. D'Agostino MA, Terslev L, Wakefield R, et al. Novel algorithms for the pragmatic use of ultrasound in the management of patients with rheumatoid arthritis: from diagnosis to remission. Ann Rheum Dis. 2016; 75: 1902-8.
6. Nordberg LB, Lillegraven S, Lie E, et al. Patients with seronegative RA have more inflammatory activity compared with patients with seropositive RA in an inception cohort of DMARD-naïve patients classified according to the 2010 ACR/EULAR criteria. Ann Rheum Dis. 2017; 76: 341-5.
7. Boutry N, Lardé A, Lapègue F, et al. Magnetic resonance imaging appearance of the hands and feet in patients with early rheumatoid arthritis. J Rheumatol. 2003; 30: 671-9.
8. Bowen CJ, Culliford D, Dewbury K, et al. The clinical importance of ultrasound detectable forefoot bursae in rheumatoid arthritis. Rheumatology (Oxford). 2010; 49: 191-2.
9. Nieuwenhuis WP, van Steenbergen HW, Mangnus L, et al. Evaluation of the diagnostic accuracy of hand and foot MRI for early Rheumatoid Arthritis. Rheumatology (Oxford). 2017; 56: 1367-77.
10. Hossain M, Makwana N. 'Not plantar fasciitis': The differential diagnosis and management of heel pain syndrome. Orthop Trauma. 2011; 25: 198-206.
11 Sarrafian SK, Kelikian AS. Tendon sheaths and bursae. In: Kelikian AS, editor. Sarrafian's anatomy of the foot and ankle: Descriptive, topographic, functional, 3rd ed. Philadelphia, PA: Wolters Kluwer, Lippincott Williams & Wilkins. 2011, 292-301.
12. Yamakado K. Subcalcaneal bursitis with plantar fasciitis treated by arthroscopy. Arthrosc Tech. 2013; 2: 135-9.
13. Kawamura K, Momohara S. Multiple rheumatoid bursal cysts with invasion into the calcaneus. J Clin Rheumatol. 2009; 15: 430.
14. Falsetti P, Frediani B, Acciai C, et al. Heel fat pad involvement in rheumatoid arthritis and in spondyloarthropathies: an ultrasonographic study. Scand J Rheumatol. 2004; 33: 327-31.
15. Falsetti P, Frediani B, Acciai C, et al. Ultrasonography and magnetic resonance imaging of heel fat pad inflammatory-oedematous lesions in rheumatoid arthritis. Scand J Rheumatol. 2006; 35: 454-8.