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Pustulotic arthro-osteitis (Sonozaky syndrome)

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A 35-year-old woman was evaluated for painful swelling of left sternoclavicular (SC) joint unresponsive to various nonsteroidal anti-inflammatory drugs (NSAIDs). Her symptoms had started 8 months prior, with gradual deterioration, resulting in awakening during night and painful lifting of left upper arm. The patient had a 5-year history of palmoplantar pustulosis (PPP) unresponsive to topical treatment, and no family history for psoriasis and rheumatic disorders. On an orthopedic evaluation 2 weeks earlier, she was suggested to start treatment with methylprednisolone 16 mg per day, present laboratory tests and magnetic resonance imaging (MRI) examination of left SC joint, and consult a rheumatologist afterward. The patient referred significant improvement of SC joint swelling and pain, and PPP with the corticosteroid treatment. On physical examination, there were some resolved pustules and little exfoliation on both palms and the left sole, and swelling of the left SC joint with mild tenderness on palpation (Figure 1), without other pathological findings. There were normal inflammatory markers (C-reactive protein = 0.08 mg dL^{-1} and erythrocyte sedimentation rate = 17 mm/1st h) and negative HLA-B27. MRI of the left SC joint showed subchondral bone marrow edema and osteitis on the manubrium of the sternum and on the sternal end of the left clavicle, and synovitis of the left SC joint (Figure 2). Based on these findings, a diagnosis of pustulotic arthro-osteitis (PAO), alternatively termed as Sonozaky syndrome, was made.

PAO, first described by Sonozaki et al.,[1] is a rare chronic inflammatory arthropathy associated with PPP, a skin disease characterized by recurrent eruptions of sterile pustules, erythema, and exfoliation, located exclusively, and often symmetrically, on the palms and/or soles. It is included in the group of seronegative spondyloarthropathies (SpA); however, there is no association with HLA-B27. Musculoskeletal involvement in PAO is most commonly seen in the SC joints. Other frequently affected regions include the spine and the sacroiliac and peripheral joints. MRI examination demonstrates bone marrow edema/osteitis, soft

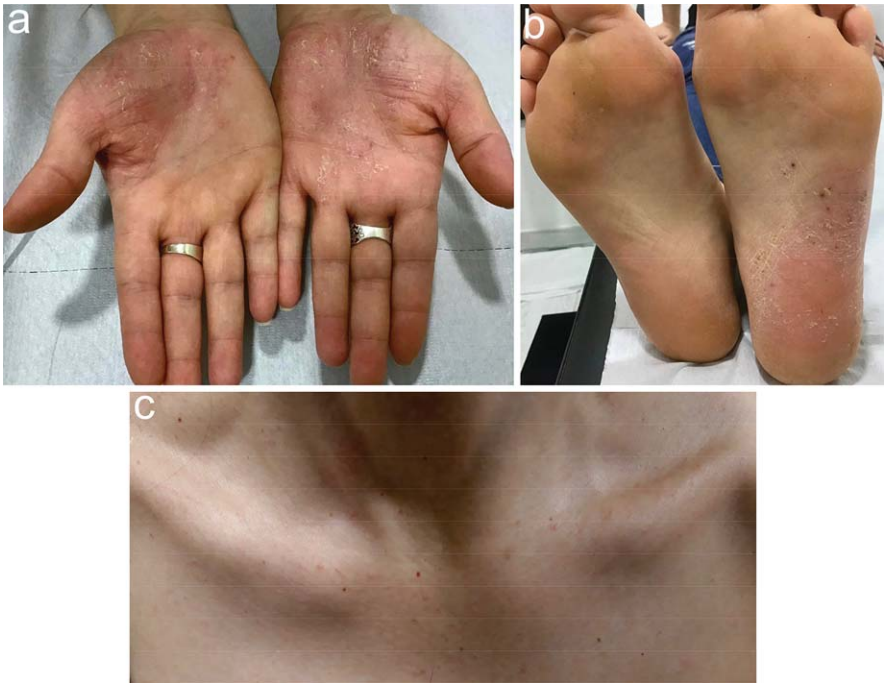

Figure 1. a-c. (a) and (b) Resolved pustules and little exfoliation on both palms and the left sole, and (c) swelling of the left sternoclavicular joint.

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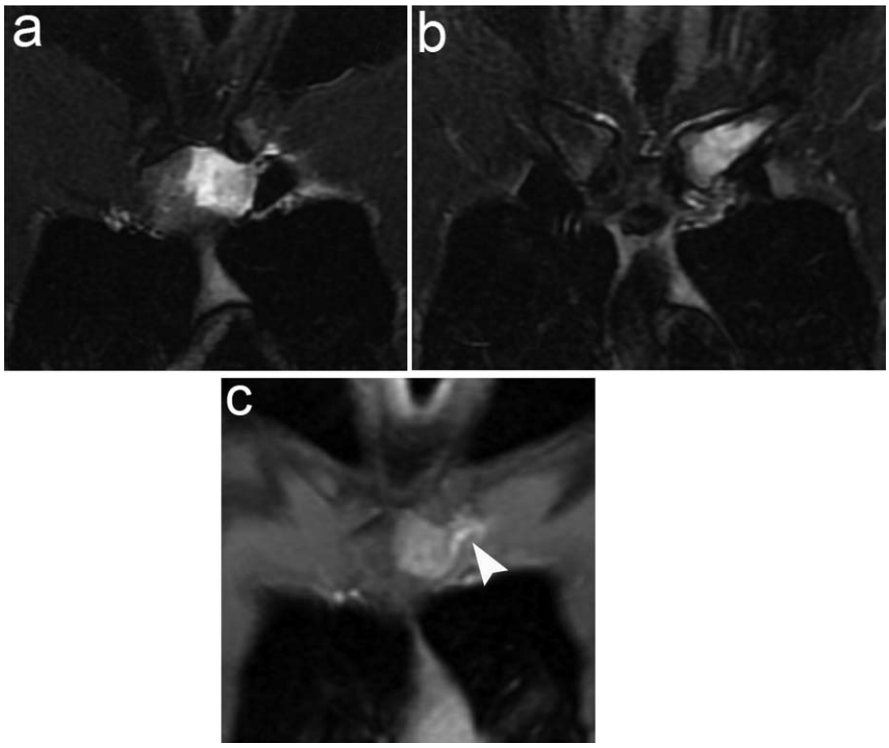

Figure 2. a-c. Coronal MRI showing (a) and (b) subchondral high signal intensity (bone marrow edema) on the manubrium of the sternum and on the sternal end of the left clavicle on STIR images, and (c) subchondral enhancement (osteitis) of the manubrium of the sternum and synovium enhancement (synovitis) of the left sternoclavicular joint (arrowhead) on fat-suppressed T1W image after gadolinium administration.

tissue swelling/edema, and bone erosion and proliferation of affected regions. PAO must be differentiated from other types of seronegative SpA such as psoriatic arthritis (PsA) and SAPHO (synovitis, acnes, pustulosis, hyperostosis, and osteitis) syndrome.[2] The SC joint involvement is uncommon in other SpA; thus, PAO differs from this group. Peripheral joint involvement is a common feature of PsA. Erosive course and polyarticular joint involvement in PsA distinguish it from PAO. Many clinical features of SAPHO syndrome are similar to PAO. Differential diagnosis is difficult, but PAO is distinguished from SAPHO syndrome by the absence of acne and intense osteomyelitic changes in imaging examination. Treatment of PAO includes removal of focal infection (teeth and tonsils), NSAIDs, corticosteroids, and disease-modifying antirheumatic drugs used for PsA, while in some refractory cases, biologic treatment has been used with encouraging results.

In conclusion, PAO is a rare entity and may be confused with other diseases causing similar symptoms. The musculoskeletal involvement only of SC joints and the finding of osteitis on MRI examination in a patient with PPP are enough to support the diagnosis of PAO.

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