

Is palmoplantar pustulosis combined with hip involvement a SAPHO syndrome?

Qiong Sun^{1,2}, Fanzhang Meng³, Naigang Chen⁴, Chen Li⁵, Zhenhua Ying^{2,6,7}

¹The Second College of Clinical Medicine, Zhejiang Chinese Medical University, Zhejiang, China

²Department of Rheumatology and Immunology, Center for General Practice Medicine, Zhejiang Provincial People's Hospital (Affiliated People's Hospital, Hangzhou Medical College), Zhejiang, China

³School of Clinical Medicine, Beijing University of Chinese Medicine, Beijing, China

⁴Department of Medical Imaging, Fangshan Hospital, Beijing University of Chinese Medicine, Beijing, China

⁵Department of Rheumatology, Fangshan Hospital, Beijing University of Chinese Medicine, Beijing, China

⁶Institute of Rheumatology and Immunology, Hangzhou Medical College, Zhejiang, China

⁷Zhejiang Provincial Key Laboratory of Traditional Chinese Medicine, Cultivation for Arthritis Diagnosis and Treatment, Zhejiang, China

Adv Dermatol Allergol 2024; XLI (3): 328–330

DOI: <https://doi.org/10.5114/ada.2024.141097>

Palmar-plantar pustulosis (PPP) is a chronic inflammatory disease characterized by aseptic pustules of erythematous keratotic lesions primarily involving the palms and soles of the hands [1]. According to the existing diagnostic criteria for SAPHO syndrome, when a patient presents with palmoplantar pustulosis combined with osteoarticular involvement, then SAPHO syndrome is diagnosed. The common sites of bone and joint involvement in SAPHO syndrome are the anterior chest wall (ACW), the midshaft bones, and the long bones of the extremities [2]. There are no reports of PPP patients with combined hip involvement diagnosed as SAPHO syndrome. Here, we report the case of PPP combined with hip involvement, which was treated with JAK inhibitors combined with bisphosphonates with significant improvement in symptoms.

A 36-year-old man with a 7-year history of left inguinal pain was treated intermittently with oral nonsteroidal anti-inflammatory drugs (NSAIDs) for pain relief. Three years ago, plantar pustules appeared, and a diagnosis of palmoplantar pustulosis (PPP) was made. The patient denied the presence of anterior chest pain during the course of the disease, the use of hormones, and the use of alcohol, tobacco, and other bad habits. Recent hip computed tomography (CT) suggested: localized cortical discontinuity and slight collapse of the left femoral head, and multiple necrotic lesions with peripheral osteosclerosis. Hip magnetic resonance imaging (MRI) compression fat sequence suggested: bone marrow oedema of the left femoral head (Figure 1). Laboratory tests showed

that HLA-B27 was negative; C-reactive protein 13 ↑ (0–10 mg/l); and blood sedimentation 25 ↑ (0–15 mm/h). The patient's pain improved after 3 days of bisphosphonate (zoledronic acid 4 mg qd) intravenous treatment, followed by oral tofacitinib 5 mg bid treatment, and the patient's symptoms of the rash improved significantly after 1 month of treatment (Figure 2).

Palmoplantar pustulosis is a rare, chronic, relapsing inflammatory disease that is the most common cutaneous manifestation of the SAPHO syndrome [3]. Osteitis and osteophytes are the core clinical manifestations of SAPHO syndrome, and when typical bone lesions are located at characteristic target sites, (e.g., sternoclavicular joints (93.0%), costovertebral joints (87.0%), sternoclavicular joints (84.5%), spine (33%), and sacroiliac joints (16.6%), among others [3]), the diagnosis of SAPHO syndrome is not difficult. However, if atypical sites are involved in patients with dermatologic disease, (e.g., mandible (2–10%) [4], skull (4.5%) [5] etc.), making the diagnosis is often challenging. The hip is a rare site of involvement in SAPHO syndrome, with a prevalence of only 3% [6].

SAPHO syndrome and spondyloarthritis all have the potential to involve hip joint lesions, and we need to identify such diseases in a timely manner. Approximately 92.9% of SAPHO and 97.7% of SpA patients with MRI pressure-lipid sequences are capable of developing bone marrow oedema and patchy bone oedema [7]. A study by Iida *et al.* found that bone marrow oedema was highly correlated with subsequent femoral head collapse and that bone marrow oedema

Address for correspondence: Zhenhua Ying, MD, Department of Rheumatology and Immunology, Center for General Practice Medicine, Zhejiang Provincial People's Hospital (Affiliated People's Hospital, Hangzhou Medical College); Institute of Rheumatology and Immunology, Hangzhou Medical College; Zhejiang Provincial Key Laboratory of Traditional Chinese Medicine, Cultivation for Arthritis Diagnosis and Treatment, Zhejiang, China, e-mail: yingzh2021@163.com; Chen Li, Department of Rheumatology, Fangshan Hospital, Beijing University of Chinese Medicine, Beijing, China, e-mail: casio1981@163.com

Received: 5.04.2024, **accepted:** 12.04.2024, **online publication:** 30.06.2024.

This is an Open Access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 International (CC BY-NC-SA 4.0) license (<http://creativecommons.org/licenses/by-nc-sa/4.0/>)

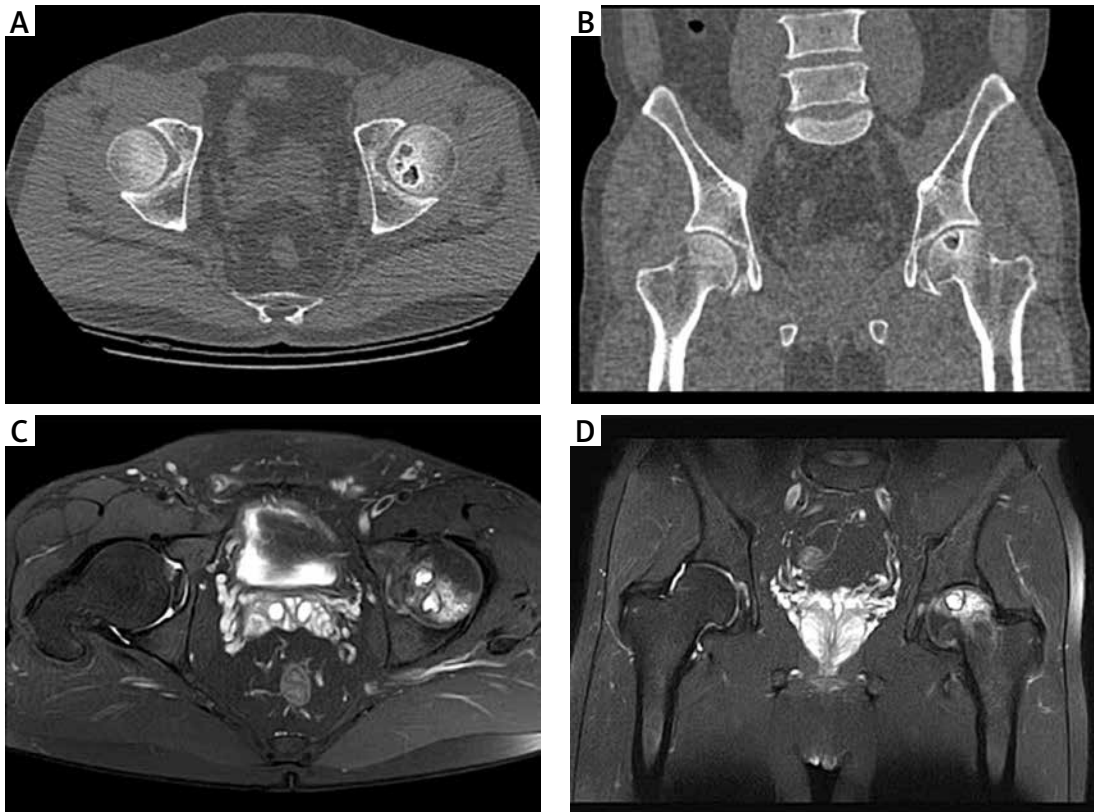


Figure 1. A, B – Hip CT suggested: localized cortical discontinuity and slight collapse of the left femoral head, and multiple necrotic lesions with peripheral osteosclerosis. **C, D** – Hip MRI compression fat sequence suggested: bone marrow oedema of the left femoral head



Figure 2. A – Prior to treatment, the patient had a scattered rash on the soles of the feet. **B** – After tofacitinib and bisphosphonate treatment, the patient's plantar lesions improved significantly

was considered a marker of potential progression to advanced osteonecrosis [8]. Some reports suggest that hip involvement in AS may be associated with synovitis or attachment point inflammation [9]. Hip MRI manifestations in patients with AS may show joint effusion and synovial enhancement due to synovitis in addition

to bone marrow oedema [10]. The MRI of the hip joint in our patient showed bone marrow oedema without HLA-B27 positivity and other AS manifestations, so we hypothesized that this patient presented with palmoplantar pustulosis combined with hip arthropathy, a special type of SAPHO syndrome.

Acknowledgments

The authors are grateful to all participants for their participation in this paper. All authors have read and approved all versions of the manuscript, its content, and its submission to the *Advances in Dermatology and Allergology/Postępy Dermatologii i Alergologii*.

Qiong Sun and Fanzhang Meng contribute equally to this letter and should be the co-first author.

Data availability

The datasets analysed for this study are available from the corresponding author Dr. Chen Li (casio1981@163.com) upon reasonable request.

Funding

This work was supported by the Zhejiang Provincial Key Laboratory of Traditional Chinese Medicine, Cultivation for Arthritis Diagnosis and Treatment (grant number: C-2023-W2083); and National Natural Science Foundation of China (grant number: 82074246).

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Conflict of interest

The authors declare no conflict of interest.

References

1. Brunasso AMG, Massone C. Recent advances in palmoplantar pustulosis. *Fac Rev* 2021; 10: 62.
2. Quirico Rodríguez M, Casáns Tormo I, Redal Peña MC, Lopez Castillo V. [The importance of bone scintigraphy in the diagnosis of SAPHO syndrome]. *Rev Esp Med Nucl* 2010; 29: 127-30.
3. Furer V, Kishimoto M, Tsuji S, et al. The diagnosis and treatment of adult patients with SAPHO Syndrome: controversies revealed in a multidisciplinary international survey of physicians. *Rheumatol Ther* 2020; 7: 883-91.
4. Wang M, Li Y, Cao Y, et al. Mandibular involvement in SAPHO syndrome: a retrospective study. *Orphanet J Rare Dis* 2020; 15: 312.
5. Oguro E, Kato Y, Takamatsu H, et al. A case of SAPHO syndrome with the lesions limited to the skull. *Rheumatol Adv Pract* 2020; 4: rkaa034.
6. Wuriliga, Wu X, Cao Y, Li C. Successful treatment of hip involvement in SAPHO syndrome with baricitinib. *Rheumatol Adv Pract* 2022; 6: rkac084.
7. Zhang LH, Han SB, Song L, et al. Comparative analysis and differentiation between SAPHO syndrome and spondylo-

- arthropathies using whole-spine MRI. *Clin Radiol* 2021; 76: 394.e9-394.e14.
8. Iida S, Harada Y, Shimizu K, et al. Correlation between bone marrow edema and collapse of the femoral head in steroid-induced osteonecrosis. *AJR Am J Roentgenol* 2000; 17: 735-43.
9. Yu C, Zhan X, Liang T, et al. Mechanism of hip arthropathy in ankylosing spondylitis: abnormal myeloperoxidase and phagosome. *Front Immunol* 2021; 12: 572592.
10. Huang ZG, Zhang XZ, Hong W, et al. The application of MR imaging in the detection of hip involvement in patients with ankylosing spondylitis. *Eur J Radiol* 2013; 82: 1487-93.