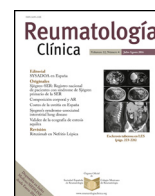




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Fever, Palmoplantar Pustules and Oligoarthritis in a Young Woman[☆]



Fiebre, lesiones vesículo-ampollosas y oligoartritis en una paciente joven

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Case Report

The patient was a 41-year-old woman, with no other remarkable findings of interest. She presented with a 3-week history of fever, arthralgia, swollen neck and pustular lesions on her hands, accompanied by pain. Over the last few days, she reported parasternal swelling that also affected right shoulder.¹ Physical exploration revealed erythema and edema in neck, glenohumeral and right sternoclavicular arthritis, and diffuse dorsopalmar pustular lesions on her hands (up to 2 mm) (Figs. 1–3), outer ear (Fig. 4) and neck and back (with acne). We found no other signs or lymphadenopathy.

Diagnosis/disease Course

Laboratory findings: leukocytosis 13,000 (normal: 10,020); platelets 68,000; C-reactive protein (CRP) 14.7 (<0.5 mg/dL); and erythrocyte sedimentation rate 20 mm/1st hour. Moreover, antinuclear antibodies, anti-DNA antibodies, antineutrophil cytoplasmic antibodies, anti-extractable nuclear antigen antibodies, angiotensin-converting enzyme, rheumatoid factor, anti-cyclic citrullinated peptide antibodies, immunoglobulins, complement and TSH/free T4 were normal. Mantoux and serology (hepatitis B and C viruses, human immunodeficiency virus, syphilis) and cultures were negative (blood culture, urine culture, exudates from pharynx, urethra and pustules).^{2–4} Neck and abdominal ultrasound and chest and shoulder radiographs provided no useful evidence. Biopsy of the skin lesions: compatible with palmoplantar pustulosis. Bone scintigraphy: increased symmetrical uptake in

sternoclavicular “bull’s head”, compatible with synovitis, acne, pustulosis, hyperostosis and osteitis (SAPHO) (Fig. 5). Treatment was begun with nonsteroidal anti-inflammatory drugs, systemic (16 mg oral methylprednisolone) and topical (betamethasone-gentamycin ointment) corticosteroids. The fever, joint involvement and skin lesions remitted (scaling; Fig. 6), and the analytical findings were normalized (previous CRP, leukocytosis and thrombocytopenia). Once corticosteroid therapy had been reduced, methotrexate was begun with a rapid dose escalation, and the response was positive and tolerance was good.^{2,3,5,6}

Discussion

The SAPHO syndrome is an uncommon disorder of unknown etiology, that affects young men and women in a similar distribution.^{6,7} Most authors classify it under the spondyloarthritides, although it could be a reactive arthropathy secondary to



Fig. 1. Diffuse pustular lesions on the dorsum of the hands.

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Fig. 2. Diffuse pustular lesions on the dorsum of the hands.



Fig. 5. Resolution of pustular lesions on the palms of the hands (scaling phase).



Fig. 3. Diffuse pustular lesions on the palms of the hands.



Fig. 4. Pustular lesion on right outer ear.



Fig. 6. Bone scintigraphy with typical sternoclavicular uptake of the so-called "bull's head", compatible with synovitis, acne, pustulosis, hyperostosis and osteitis (SAPHO) syndrome.

an infection by an agent with a low virulence.^{2,4,7,8} On occasion, it may prove difficult to diagnose because it overlaps with other infectious conditions that present with fever and skin lesions.^{8,9} The presence of edema in neck and chest, acne and oligoarthritis,

in the absence of bacteriological isolation, leads us to suspect an autoimmune etiology.¹⁰ The skin biopsy and scintigraphic data subsequently confirmed our presumed diagnosis.

Ethical Disclosures

Protection of human and animal subjects. The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

Conflicts of Interest

The authors declare they have no conflicts of interest.

References

1. Nanba F, Hayashi T, Ishiga M, Kishimoto M, Yagi S, Okimoto N. Case of SAPHO syndrome with significant sternocostoclavicular hyperostosis [Article in Japanese]. *Nihon Kokyuki Gakkai Zasshi.* 2010;48:810–4.
2. Gharsallah I, Souissi A, Dhahri R, Boussetta N, Sayeh S, Métoui L, et al. SAPHO syndrome [Article in French]. *Rev Med Interne.* 2014;35:595–600.
3. Rosero A, Ruano R, Martín M, Hidalgo C, García-Talavera J. Acute venous thrombosis as complication and clue to diagnose a SAPHO syndrome case. A case report. *Acta Reumatol Port.* 2013;38:203–6.
4. Yamamoto T. Pustulotic arthro-osteitis associated with palmoplantar pustulosis. *J Dermatol.* 2013;40:857–63.
5. Kim CH, Kadhim S, Julien C. Treatment of pain in SAPHO (synovitis, acne, pustulosis, hyperostosis, and osteitis) syndrome. *PM R.* 2014;6:92–5.
6. Nguyen MT, Borchers A, Selmi C, Naguwa SM, Cheema G, Gershwin ME. The SAPHO syndrome. *Semin Arthritis Rheum.* 2012;42:254–65.
7. Carranco-Medina TE, Hidalgo-Calleja C, Calero-Paniagua I, Sánchez-González MD, Quesada-Moreno A, Usategui-Martín R, et al. Thrombotic manifestations in SAPHO syndrome. Review of the literature [Article in English, Spanish]. *Reumatol Clin.* 2015;11:108–11.
8. Carneiro S, Sampaio-Barros PD. SAPHO syndrome. *Rheum Dis Clin North Am.* 2013;39:401–18.
9. Kundu BK, Naik AK, Bhargava S, Srivastava D. Diagnosing the SAPHO syndrome: a report of three cases and review of literature. *Clin Rheumatol.* 2013;32:1237–43.
10. Zuo RC, Schwartz DM, Lee CC, Anadkat MJ, Cowen EW, Naik HB. Palmoplantar pustules and osteoarticular pain in a 42-year-old woman. *J Am Acad Dermatol.* 2015;72:550–3.