

Bone Scan and SPECT/CT Scan in SAPHO Syndrome

Abstract

Synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome is a rare clinical entity involving musculoskeletal and dermatologic systems. Its main features are prominent inflammatory cutaneous and articular manifestations. Anterior chest wall pain, more commonly at sternoclavicular and sternocostal joints, along with palmoplantar pustulosis and acne can point to this uncommon syndrome. Here, we report a case of a 15-year-old female, who presented with swelling at the anterior chest wall, was referred for the bone scan. Bone scintigraphy and single-photon emission computed tomography (SPECT/CT) detected classical "bull-head" sign, and further physical examination, for skin lesions, confirmed the diagnosis.

Keywords: Acne, bone scan, hyperostosis and osteitis syndrome, pustulosis, single-photon emission computed tomography-computed tomography, synovitis

A 15-year-old female with a history of pain and swelling in the anterior chest wall, centered at bilateral clavicular regions for 8 months, was referred for bone scintigraphy. A complete blood count, erythrocyte sedimentation rate, and serum C-reactive protein were normal. Whole-body bone scan anterior and posterior images [Figure 1a and b] revealed increased tracer uptake in bilateral sternoclavicular joints (left >> right), left clavicle, sternum, bilateral first sternocostal joints and bilateral sacro-iliac joints. The images show intense tracer uptake at the sternoclavicular joints and sternum, which represent a "bullhead" sign, described in synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome. It was first described by Freyschmidt and Sternberg.^[1] Single-photon emission computed tomography-computed tomography (SPECT-CT) scan was also done for further evaluation. Computed tomographic three-dimensional reconstruction [Figure 1c] of the bones of the anterior chest wall showed hyperostosis of the left clavicle. Fused SPECT-CT coronal and axial images [Figure 1d-g] showed expansile osteolytic, sclerotic destructive lesions in bilateral clavicles and chondral ends of bilateral first ribs (left >> right). The characteristic "bullhead" sign raises suspicion of SAPHO syndrome. On

physical examination, rashes were detected on the palms and soles of the patient. The diagnosis of SAPHO syndrome was considered based on the presence of one of the diagnostic criteria formulated by Benhamou *et al.*^[2]

The acronym SAPHO was introduced in 1987.^[3] Few case reports of SAPHO syndrome have been discussed in the literature.^[4-8] Osteoarticular manifestations involve osteitis, hyperostosis, synovitis, arthropathy, and enthesopathy that present with pain, tenderness, and sometimes swelling over the affected areas and fever. Classical cutaneous lesions seen in SAPHO patients include palmoplantar pustulosis and severe acne.^[9] Skin lesions may vary in severity and may precede (in 50% of the cases), follow, or occur simultaneously with the onset of arthritis.^[10] The present case highlights the importance of bone scan, SPECT-CT imaging, and physical examination to produce a definite diagnosis of SAPHO.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be

How to cite this article: Gupta N, Verma R, Belho ES. Bone scan and SPECT/CT scan in SAPHO syndrome. *Indian J Nucl Med* 2019;34:349-50.

**Nitin Gupta,
Ritu Verma,
Ethel Shangne
Belho**

*Department of Nuclear
Medicine and PET/CT, Mahajan
Imaging Centre, Sir Ganga Ram
Hospital, New Delhi, India*

Address for correspondence:

*Dr. Nitin Gupta,
Department of Nuclear
Medicine and PET/CT, Mahajan
Imaging Centre, Sir Ganga Ram
Hospital, New Delhi - 110 060,
India.*

E-mail: drnitingpt@gmail.com

Access this article online

Website: www.ijnm.in

DOI: 10.4103/ijnm.IJNM_139_19

Quick Response Code:



This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

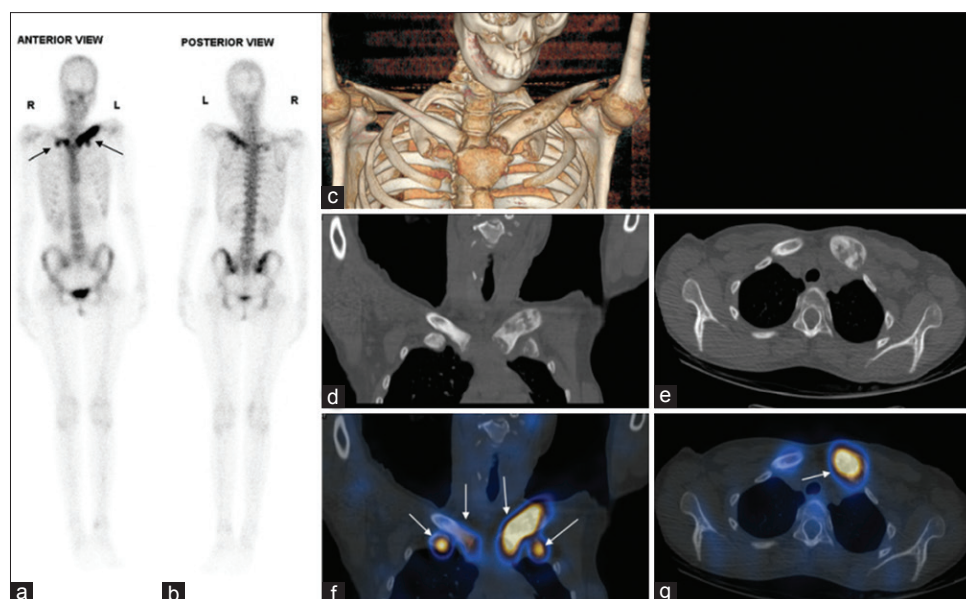


Figure 1: Whole-body bone scan anterior and posterior images (a and b) show increased tracer uptake in bilateral sternoclavicular joints (left >> right), left clavicle, sternum, bilateral first sternocostal joints, and bilateral sacroiliac joints. Computed tomographic three-dimensional reconstruction (c) of the bones of the anterior chest wall shows hyperostosis of the left clavicle. Fused single-photon emission computed tomography-computed tomography coronal and axial images (d-g) show expansile osteolytic, sclerotic destructive lesions in bilateral clavicles and chondral ends of bilateral first ribs (left >> right)

made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Freyschmidt J, Sternberg A. The bullhead sign: Scintigraphic pattern of sternocostoclavicular hyperostosis and pustulotic arthroosteitis. *Eur Radiol* 1998;8:807-12.
2. Benhamou CL, Chamot AM, Kahn MF. Synovitis-Acne-Pustulosis Hyperostosis-Osteomyelitis syndrome (SAPHO). A new syndrome among the spondyloarthropathies? *Clin Exp Rheumatol* 1988;6:109-12.
3. Chamot AM, Benhamou CL, Kahn MF, Beranek L, Kaplan G, Prost A, *et al.* [Acne-pustulosis-hyperostosis-osteitis syndrome. Results of a national survey 85 cases]. *Rev Rhum Mal Osteoartic* 1987;54:187-96.
4. Canbaz F, Gonullu G, Baris S, Selcuk MB, Bicakci N. SAPHO syndrome without dermatologic manifestations: Multifocal uptake mismatch on ^{99m}Tc -MDP and ^{18}F -FDG-PET/CT imaging. *Hell J Nucl Med* 2010;13:73-5.
5. Abuhid IM, Silva LC, Martins GP, de Rezende NA. Diagnosing SAPHO syndrome in suspected metastatic bone tumors. *Clin Nucl Med* 2010;35:172-4.
6. İlgen U, Turan S, Emmungil H. Bull's head sign in a patient with SAPHO syndrome *Balkan Med J* 2019;36:139-40.
7. Song X, Sun W, Meng Z, Gong L, Tan J, Jia Q, *et al.* Diagnosis and treatment of SAPHO syndrome: A case report. *Exp Ther Med* 2014;8:419-22.
8. Seven B, Varoglu E, Erdem T, Atasoy M. Bone scintigraphy results in a patient with sapho syndrome. *Eurasian J Med* 2008;40:157.
9. Monsour PA, Dalton JB. Chronic recurrent multifocal osteomyelitis involving the mandible: Case reports and review of the literature. *Dentomaxillofac Radiol* 2010;39:184-90.
10. Hayem G, Bouchaud-Chabot A, Benali K, Roux S, Palazzo E, Silbermann-Hoffman O, *et al.* SAPHO syndrome: A long-term follow-up study of 120 cases. *Semin Arthritis Rheum* 1999;29:159-71.