



Universiteit
Leiden
The Netherlands

Chronic non-bacterial osteomyelitis in SAPHO syndrome complicated by subclavian vein obstruction

Leerling, A.T.; Cañete, A.N.; Winter, E.M.

Citation

Leerling, A. T., Cañete, A. N., & Winter, E. M. (2023). Chronic non-bacterial osteomyelitis in SAPHO syndrome complicated by subclavian vein obstruction. *Rheumatology*, 62(12), E355-E356. doi:10.1093/rheumatology/kead264

Version: Publisher's Version

License: [Creative Commons CC BY-NC 4.0 license](https://creativecommons.org/licenses/by-nc/4.0/)

Downloaded from: <https://hdl.handle.net/1887/3721263>

Note: To cite this publication please use the final published version (if applicable).



Clinical vignette

Chronic non-bacterial osteomyelitis in SAPHO syndrome complicated by subclavian vein obstruction

Anne T. Leerling ^{1,2,*}, Ana Navas Cañete ^{2,3}, Elizabeth M. Winter ^{1,2}

¹Department of Internal Medicine, Division of Endocrinology, Leiden University Medical Center, Leiden, The Netherlands

²Center for Bone Quality, Leiden University Medical Center, Leiden, The Netherlands

³Department of Radiology, Leiden University Medical Center, Leiden, The Netherlands

*Correspondence to: Anne Tamar Leerling, Department of Internal Medicine, Division of Endocrinology, Center for Bone Quality, Leiden University Medical Center, Postal Zone B2-R, Albinusdreef 2, 2333 ZA Leiden, The Netherlands. E-mail: a.t.leerling@lumc.nl

A 50-year-old woman presented to our tertiary center with chest pain that had been present for 2 years, without weight loss or fever. Physical examination revealed a bony manubrial swelling with prominent vasculature, with oedema of the left hemithorax, and a pustular rash on the foot soles. ESR was 65 mm/h, haemoglobin 7.8 mmol/l, and ALP 69 U/l. Single-photon emission CT demonstrated sclerosis, hyperostosis and increased uptake of the manubrium and upper ribs (Fig. 1, left pane). The pustular rash was diagnosed as pustulosis palmo-plantaris, and the total clinical picture was suggestive of chronic non-bacterial osteomyelitis in the context of synovitis, acne, pustulosis, hyperostosis, osteitis syndrome (CNO/SAPHO). Contrast-CT showed obstruction of the left subclavian vein with expansive collaterals (Fig. 1, right pane), presumably resulting from a thrombotic event caused by hyperostotic compression. No malignant lesions were observed. Prophylactic apixaban was started, and the patient

was treated with 3-monthly i.v. pamidronate with good clinical effect on bone pain.

CNO/SAPHO mostly manifests as osteitis of the anterior chest wall in adults and often co-occurs with dermatologic or articular inflammation [1]. In long-existing CNO/SAPHO, hyperostosis may cause neurovascular compression, which may trigger thrombosis warranting anticoagulants [2].

Data availability

Data are available on request.

Funding

No specific funding was received from any bodies in the public, commercial or not-for-profit sectors to carry out the work described in this article.

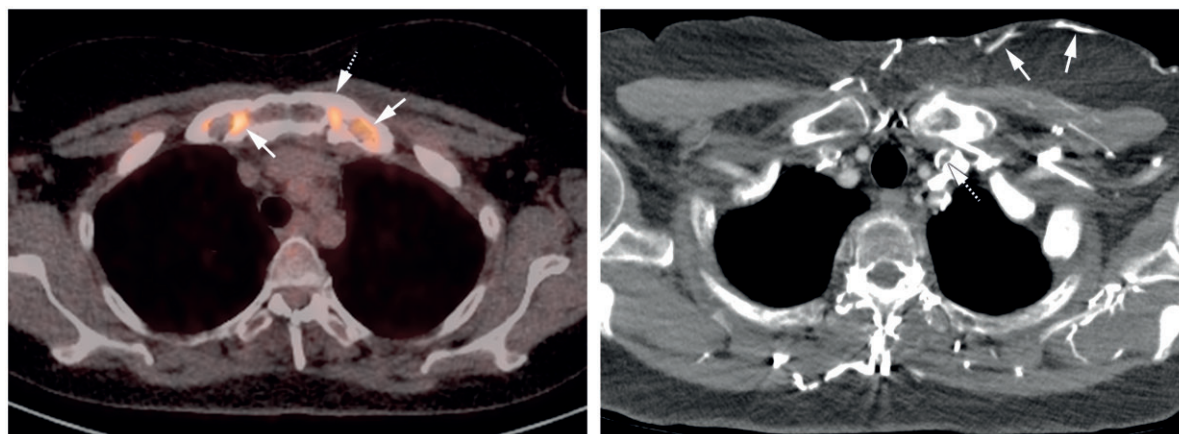


Figure 1. Single-photon emission CT and contrast CT of the anterior chest wall demonstrating CNO/SAPHO complicated by subclavian vein obstruction. Left pane: arrows indicate sclerosis, hyperostosis and increased uptake of the manubrium and upper ribs. Right pane: arrows indicate obstruction of the left subclavian vein with expansive collaterals

Disclosure statement: The authors have declared no conflicts of interest.

References

1. Leerling A, Dekkers O, Appelman-Dijkstra N, Winter E. Clinical and therapeutic diversity in adult chronic nonbacterial osteomyelitis (CNO) of the sternocostoclavicular region: a meta-analysis. *Rheumatology (Oxford)* 2023;62:512–22.
2. Godot A, Fakih O, Prati C, Wendling D, Verhoeven F. SAPHO syndrome and subclavian thrombosis: simple fortuitous association? *Eur J Intern Med* 2020;72:103–5.