

DATE: 12/04/2024

*World Journal of Rheumatology*

Dear reviewer:

Please find enclosed our manuscript entitled “Synovitis, Acne, Pustulosis, Hyperostosis, and Osteitis (SAPHO) Syndrome as a cause of Pneumothorax: A Case Report,” which we request you to consider for publication as a *Case Report* in *World Journal of Rheumatology*.

Based on the suggestions you have made regarding the manuscript, I will respond to each one individually:

1. I have not seen a list of your key words. I would recommend SAPHO syndrome, Pneumothorax, Case report.

Firstly, we sincerely appreciate you pointing out our oversights. Based on your

recommendations, we have set up the keywords: SAPHO syndrome, Pneumothorax, Case report, Methotrexate, Closed thoracic drainage.

2. Instead of writing 'Dear Editor' at the start, you could use 'Introduction' because what you are writing about in those first paragraphs is an introduction to the case.

Thank you again for correcting the format of our article. Following your suggestion, we have replaced "Dear Editor" with "Introduction".

3. The entire manuscript is the case report, according to my understanding. So instead of using 'Case report', you can use case presentation.

Thank you again for correcting the format of our article. Following your suggestion, we have replaced "Case report" with "case presentation".

4. The opening statement to your case presentation is really lacking. You could re-phrase like; 44 year old male, known hypertensive for 10years on....., also known with SAPHO syndrome, which was managed with methotrexate for one year. He had been off methotrexate for 2years. He presented with..... This is because you are not diagnosing the SAPHO syndrome now, it was already there. Only tell us the symptoms he came with this time round.

Thank you for your guidance on the language aspects. As a result, we have made revisions to the content of our article: A 44-year-old male, hypertensive for 10 years, presented with palmoplantar pustulosis and anterior chest wall pain lasting 2 years. Technetium-99m methylene diphosphonate ( $^{99m}\text{Tc}$ -MDP) bone scintigraphy revealed radioactive element accumulation in the bilateral sternoclavicular joints (Figure 1A). Based on his symptoms and imaging findings, the patient was diagnosed with SAPHO syndrome. He was treated with methotrexate for 1 year, which relieved his skin and joint pain, after which the treatment was discontinued.

5. What were the examination findings for such a patient who came in with anterior chest wall pain and dyspnea? Was the chest tender to palpation, what was the oxygen saturation, how was the chest percussion, how was the air entry in the chest?? In other words, on examination, what features pointed to your team that this patient had a pneumothorax? It would be important for readers to know how you came to that conclusion

Thank you for raising questions about the physical examination of our patient:He presented with right-sided chest pain and tenderness on chest palpation, and his oxygen saturation was 94%. And based on the patient's subsequent CT scan results, we have concluded that the patient has pneumothorax.

6. You could put a heading before the paragraph that begins with 'The occurrence of pulmonary.....' For example, 'Case Discussion'

Thank you very much for your guidance on the structure of our article. Following your suggestions, we have added the appropriate headings "Case Discussion" to this section.

7. In your discussion, could you try to explain what could cause pneumothorax in a patient with SAPHO syndrome.

Certainly, we are more than happy to engage in this discussion.SAPHO syndrome has been proposed as a transitional stage between ankylosing spondylitis and psoriatic arthritis. One study reported a 0.29% incidence of spontaneous pneumothorax in patients with ankylosing spondylitis, higher than in the general population.Additionally, 30% of patients with SAPHO syndrome test positive for HLA-B27, an antigen linked to ankylosing spondylitis. Symptomatically, some

patients with SAPHO syndrome meet diagnostic criteria for ankylosing spondylitis, with sternoclavicular joint involvement and paravertebral ossifications resembling syndesmophytes in ankylosing spondylitis. These features suggest a possible link between SAPHO syndrome and ankylosing spondylitis.

8. On the images, could you use arrows and point to what you want people to see? for example the pneumothorax on the chest CT scan.

We are delighted that you have provided guidance on our images. In response to your recommendations, we have added arrows of different colors at the appropriate locations to assist readers in better understanding and learning from our materials.

9. Could you please add DOI links to your references so that they are easily accessible to other readers as well? I could not access reference 4, so maybe if you put the DOI link it would be easier.

We apologize for the inconvenience caused by the formatting of our references. We have revised the formatting and inserted the references back into the article. Here is the specific situation:

[1] Chamot AM, Benhamou CL, Kahn MF, Beraneck L, Kaplan G, Prost A. Le syndrome acné pustulose hyperostose ostéite (SAPHO). Résultats d'une enquête nationale. 85 observations [Acne-pustulosis-hyperostosis-osteitis syndrome. Results of a national survey. 85 cases]. Rev Rhum Mal Osteoartic. 1987 Mar;54(3):187-96. French. PMID: 2954204.

[2] Bintcliffe OJ, Hallifax RJ, Edey A, Feller-Kopman D, Lee YC, Marquette CH, Tschopp JM, West D, Rahman NM, Maskell NA. Spontaneous pneumothorax: time to rethink management? Lancet Respir Med. 2015 Jul;3(7):578-88. doi: 10.1016/S2213-2600(15)00220-9. PMID: 26170077.

[3] Kurata I, Tsuboi H, Terasaki M, Shimizu M, Toko H, Honda F, Ohyama A, Yagishita M, Osada A, Ebe H, Kawaguchi H, Takahashi H, Hagiwara S, Asashima H, Kondo Y, Matsumoto I, Sumida T. Effect of biological disease-modifying anti-rheumatic drugs on airway and interstitial lung disease in patients with rheumatoid arthritis. Intern Med. 2019 Jun 15;58(12):1703-1712. doi:

10.2169/internalmedicine.2226-18. Epub 2019 Feb 25. PMID: 30799358; PMCID: PMC6630137.

[4] Dumolard A, Gaudin P, Juvin R, Bost M, Peoc'h M, Phelip X. SAPHO syndrome or psoriatic arthritis? A familial case study. *Rheumatology (Oxford)*. 1999 May;38(5):463-7. doi: 10.1093/rheumatology/38.5.463. PMID: 10371287.

[5] Li C, Liu S, Sui X, Wang J, Song W, Xu W, Xu KF, Tian X, Zhang W. Pulmonary high-resolution computed tomography findings in patients with synovitis, acne, pustulosis, hyperostosis and osteitis syndrome. *PLoS One*. 2018 Dec 5;13(12):e0206858. doi: 10.1371/journal.pone.0206858. PMID: 30517110; PMCID: PMC6281176.

[6] Colina M, Govoni M, Orzincolo C, Trotta F. Clinical and radiologic evolution of synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome: a single center study of a cohort of 71 subjects. *Arthritis Rheum*. 2009 Jun 15;61(6):813-21. doi: 10.1002/art.24540. PMID: 19479702.

[7] Hayem G, Bouchaud-Chabot A, Benali K, Roux S, Palazzo E, Silbermann-Hoffman O, Kahn MF, Meyer O. SAPHO syndrome: a long-term follow-up study of 120 cases. *Semin Arthritis Rheum*. 1999 Dec;29(3):159-71. doi: 10.1016/s0049-0172(99)80027-4. PMID: 10622680.

[8] Danve A. Thoracic manifestations of ankylosing spondylitis, inflammatory bowel disease, and relapsing polychondritis. *Clin Chest Med*. 2019 Sep;40(3):599-608. doi: 10.1016/j.ccm.2019.05.006. Epub 2019 Jul 6. PMID: 31376894.

[9] Maugars Y, Berthelot JM, Ducloux JM, Prost A. SAPHO syndrome: a followup study of 19 cases with special emphasis on entheses involvement. *J Rheumatol*. 1995 Nov;22(11):2135-41. PMID: 8596157.

[10] Shen K, Yang CL, Yin G, Xie QB. Sacroiliitis and spondylitis with sternoclavicular hyperostosis: SAPHO or an ankylosing spondylitis variant? *Chin Med J (Engl)*. 2016 Jan 5;129(1):110-1. doi: 10.4103/0366-6999.172607. PMID: 26712445; PMCID: PMC4797530.

Lastly, we express our sincere gratitude for your astute observations on the issues in our paper, which have underscored the importance of thorough preparation and accurate information when reporting a case related to the respiratory system. And considering the occurrence of pneumothorax one year after discontinuing the medication, we are inclined to view this as part of the natural course of the disease rather than a direct result of the previous drug therapy. These analyses and discussions will guide us to be more cautious and comprehensive in handling similar cases in the future, ensuring the most accurate diagnoses and treatment recommendations are provided.

Sincerely,

Chen Li

Fangshan Hospital, Beijing University of Chinese Medicine, Beijing, China

Address: No.4, Chengguan Health Road, Fangshan District, Beijing, China

E-mail:casio1981@163.com

ORCID: <https://orcid.org/0000-0002-8527-1680>

YuanHao Wu, First Teaching Hospital of Tianjin University of Traditional Chinese Medicine, Tianjin, China

Address: Changling Road, Xiqing District, Tianjin, China

E-mail:doctor.wuyh@gmail.com