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CASE REPORT

Synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome as a cause of pneumothorax: A case report

Zi-Xiang Zheng, Meng-Jiao Gu, Tian-Lun Kang, Yu-Ru Zhang, Yu-Nuo Wang, Chen Li, Yuan-Hao Wu

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Abstract

BACKGROUND

Synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome, a rare inflammatory disease, was described in 1987 by Chamot *et al*, who identified its common characteristics and transitional forms through case reports. The incidence rate is now 1 in 10000 among Caucasians and 0.00144 in 10000 among Japanese. Pneumothorax occurs when gas enters the pleural cavity, typically due to lung disease or chest trauma, and is characterized as either spontaneous or traumatic. Spontaneous pneumothorax is commonly linked to congenital lung tissue abnormalities or lung conditions, such as ruptured pulmonary bullae, emphysema, and bronchial asthma. Respiratory involvement in SAPHO syndrome has historically been noted.

CASE SUMMARY
We present the case of a 44-year-old male patient who had previously been diagnosed with SAPHO syndrome. Following a two-year cessation of methotrexate therapy, he experienced recurrent anterior chest wall and joint pain accompanied by dyspnea. We conducted a comprehensive examination for the patient, which included physical examination, laboratory tests, and imaging studies. The clinical presentation and treatment outcomes suggest that his pneumothorax was related to an episode of SAPHO syndrome. We performed closed thoracic drainage surgery for the patient and treated his symptoms with etoricoxib (60 mg daily). Upon discharge, methotrexate was prescribed again. In subsequent follow-ups, there was no recurrence of joint and respiratory symptoms observed in the patient.

CONCLUSION
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 linked between SAPHO syndrome and ankylosing spondylitis.

Key Words: Synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome; Pneumothorax; Methotrexate; Closed thoracic drainage; Case report

Core Tip: This case report introduces a novel association between synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome and pneumothorax. The 44-year-old patient's diagnosis and treatment underscore the importance of recognizing this rare but serious condition, broadening our understanding of SAPHO syndrome's clinical manifestations.

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INTRODUCTION

Synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome, a rare inflammatory disease, was described in 1987 by Chamot *et al*[1] who identified its common characteristics and transitional forms through case reports. The incidence rate is now 1 in 10000 among Caucasians and 0.00144 in 10000 among Japanese[2].

Pneumothorax occurs when gas enters the pleural cavity, typically due to lung disease or chest trauma, and is characterized as either spontaneous or traumatic[3]. Spontaneous pneumothorax is commonly linked to congenital lung tissue abnormalities or lung conditions, such as ruptured pulmonary bullae, emphysema, and bronchial asthma. Respiratory involvement in SAPHO syndrome has historically been noted. In this report, we present a case of a 44-year-old man who developed pneumothorax following SAPHO syndrome diagnosis.

CASE PRESENTATION

Chief complaints

The patient reported recurrent anterior chest wall and joint pain accompanied by dyspnea for thirteen days.

History of present illness

Two years after discontinuing methotrexate use, he experienced recurrent anterior chest wall and joint pain accompanied by dyspnea. He denied a history of coronary heart disease, diabetes, chronic obstructive pulmonary disease, tuberculosis, and smoking. He presented with right-sided chest pain and tenderness on chest palpation, and his oxygen saturation was 94%.

History of past illness

The patient, hypertensive for 10 years, presented with palmoplantar pustulosis and anterior chest wall pain lasting 2

years. Technetium-99m methylene diphosphonate (^{99m}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.

Personal and family history

He denied a history of coronary heart disease, diabetes, chronic obstructive pulmonary disease, tuberculosis, and smoking.

Physical examination

He presented with right-sided chest pain and tenderness on chest palpation, and his oxygen saturation was 94%.

Laboratory examinations

Tuberculosis was ruled out with a negative T-SPOT test. Blood tests indicated an elevated C-reactive protein level at 2.24 mg/dL (normal: < 0.3 mg/dL), whereas serum procalcitonin was negative, excluding bacterial infection.

Imaging examinations

Based on the patient's symptoms and computed tomography (CT) imaging, he was diagnosed with right-sided pneumothorax (Figure 1B), and a chest radiograph revealed 37% lung collapse. The patient underwent closed thoracic drainage surgery, which alleviated his dyspnea, and follow-up chest CT one week later confirmed no pneumothorax (Figure 1C).

FINAL DIAGNOSIS

The clinical presentation and treatment outcomes suggest that his pneumothorax was related to an episode of SAPHO syndrome.

TREATMENT

The patient underwent closed thoracic drainage surgery, which alleviated his dyspnea, and follow-up chest CT one week later confirmed no pneumothorax (Figure 1C). Subsequent treatment with etoricoxib (60 mg daily) relieved his anterior chest wall and joint pain.

OUTCOME AND FOLLOW-UP

Following the discontinuation of methotrexate two years prior, the patient resumed the medication regimen, and in our subsequent follow-up, no recurrence of joint and respiratory symptoms was observed.

DISCUSSION

The occurrence of pulmonary and pleural lesions in SAPHO syndrome is not uncommon. The first reported case of SAPHO syndrome complicated by pleural effusion appeared in 1999[4]. In a previous study evaluating high-resolution CT findings in the lungs of patients with SAPHO syndrome, irregular linear opacities were observed in 29 (43.3%) patients, opacities in 22 (32.8%), ground-glass opacities in 11 (16.4%), pleural thickening in 9 (13.4%), solitary nodules in 6 (9%), bronchiectasis in 3 (4.5%), pulmonary bulla in 2 (3%), multiple nodules in 1 (1.5%), and reticular patterns in 1 (1.5%) [5]. However, a literature search on respiratory manifestation in patients with SAPHO syndrome did not identify any previously reported cases involving pneumothorax or its symptoms.

SAPHO syndrome has been proposed as a transitional stage between ankylosing spondylitis and psoriatic arthritis[6, 7]. One study reported a 0.29% incidence of spontaneous pneumothorax in patients with ankylosing spondylitis, higher than in the general population[8]. Additionally, 30% of patients with SAPHO syndrome test positive for HLA-B27[9], 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[10]. These features suggest a possible linked between SAPHO syndrome and ankylosing spondylitis. The present case report is the first to document the coexistence of SAPHO syndrome and pneumothorax.

CONCLUSION

This case report underscores a novel association between SAPHO syndrome and spontaneous pneumothorax, previously

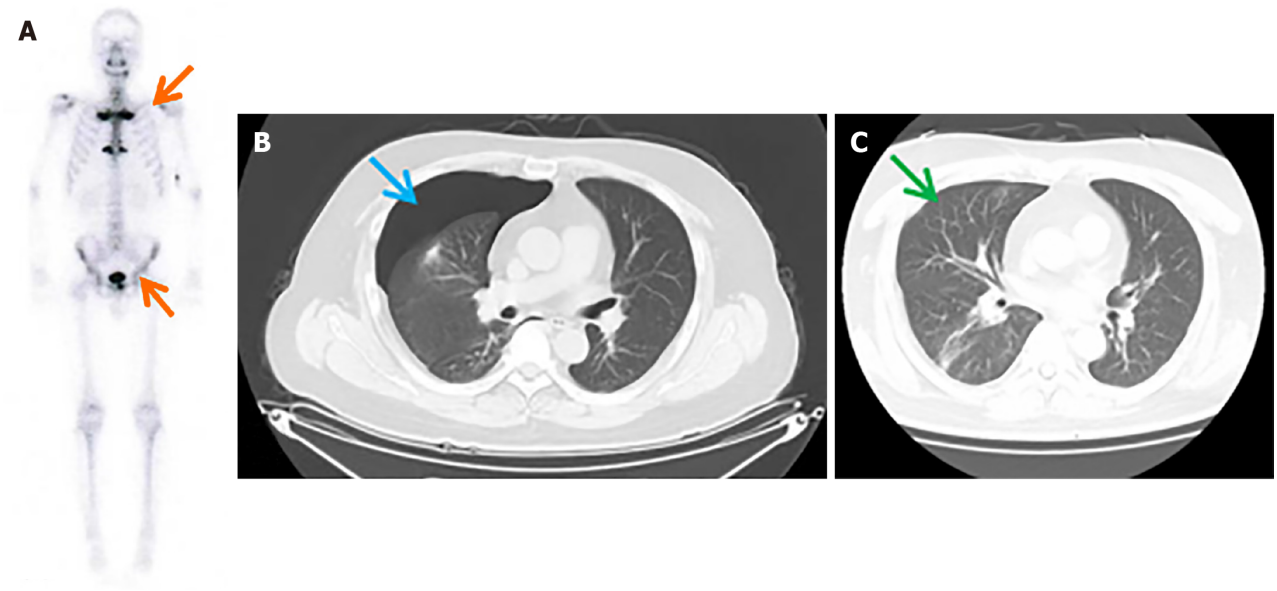

Figure 1 ^{99m}Tc-MDP bone scintigraphy and pneumothorax images in a patient with synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome. A: ^{99m}Tc-MDP bone scintigraphy revealing abnormal radioactivity accumulation in the anterior chest wall and bilateral sternoclavicular joints (indicated by the orange arrows); B: Computed tomography scan showing a right-sided pneumothorax (indicated by the blue arrow); C: Follow-up chest radiograph, taken one week after completion of pneumothorax treatment, showing resolution (indicated by the green arrow).

unreported. The patient's recovery following treatment and the absence of symptom recurrence upon methotrexate resumption suggest a potential link between SAPHO syndrome and pneumothorax development. This observation warrants further investigation into the relationship between these conditions and the need for vigilant monitoring of respiratory issues in SAPHO syndrome patients.

FOOTNOTES

Author contributions: Zheng ZX and Gu MJ collected the case content and wrote the manuscript; Kang TL and Zhang YR provided guidance on manuscript writing; Wang YN was responsible for finding relevant materials; Li C and Wu YH jointly proposed the overall direction of the topic. All authors have read and approved the final manuscript.

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