P-25 Successful treatment of plural effusion with glucocorticoid in a patient with Synovitis-acne-pustulosis- hyperostosis-osteitis syndrome

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Introduction: Synovitis-acne-pustulosis-hyperostosis-osteitis (SAPHO) syndrome is a rare inflammatory disorder in which bone, joints and skin are mainly affected. Here we report a case of pleural effusion due to SAPHO syndrome, which was successfully treated with glucocorticoid.

Case Presentation: A 57-year-old woman with a history of palmoplantar pustulosis(PPP) developed anterior chest pain and dyspnea two weeks prior to admission. She visited a nearby clinic and chest X-ray showed bilateral pleural effusions. She was referred to our hospital. Vital signs showed blood pressure 144/105 mmHg, pulse rate 89/min, temperature 35.9℃, respiration rate 20/min, and SpO2 97% on room air. Breath sounds were diminished in both lower lung fields. There was tenderness at the sternocostal joints. Desquamation was observed in her palms and soles. Labs showed mild elevation of leukocyte count and prolonged erythrocyte sedimentation rate. Liver and kidney functions were normal. Pleural fluid analysis showed exudative effusions and lymphocytosis. Adenosine deaminase was low. Cytology, smear and culture of pleural effusion were negative. Blood cultures were negative. Bone scintigraphy revealed accumulation at the sternum. Given her history of PPP and the findings of sterile osteitis of the sternum, clinical diagnosis of SAPHO syndrome was made. After excluding other possibilities, SAPHO syndrome was assumed to be the cause of the pleural effusion. NSAID showed no improvement. Oral prednisolone 60 mg daily was commenced. Thereafter, pleural effusion disappeared. She is currently followed at an outpatient clinic.

Discussion: Our case was unique because of the presence of pleural effusion. We searched English and Japanese articles and only few cases have been reported. Physicians should consider the possibility of SAPHO syndrome in patients with anterior chest pain and pleural effusions.