Clinical vignette

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Bull's head sign in the scintigraphy of a young female with recurrent chest pain

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Abstract

In this case we report a 38-year-old female patient with history of recurrent retrosternal chest pain lasting almost 5 years. Standard X-rays of chest and spine revealed no abnormalities. In a physical examination tenderness of anterior chest wall was observed, especially in sternoclavicular areas. SAPHO (synovitis, acne, pustulosis, hyperostosis, osteitis) syndrome was taken into consideration despite of lack of typical skin lesions (acne, pustulosis). We decided to implement [^{99m}Tc]Tc-MDP scintigraphy. Increased osteoblastic activity (intense [^{99m}Tc]Tc-MDP) in manubriosternal and both sternoclavicular regions represents bull's head sign which is a rare finding, but pathognomonic to SAPHO syndrome. After a 3-month therapy with aceclofenac 100 mg, total remission was reached. If we rule out this rare condition like SAPHO based on lack of abnormalities in X-rays, the reason of symptoms could be still unrecognized. [^{99m}Tc]Tc-MDP scintigraphy is valuable to show even subclinical areas of involvement and to monitor treatment response in SAPHO syndrome. This case proved significant role of whole body scintigraphy to make diagnosis of SAPHO syndrome in patients with non-cardiac chest pain and lack of abnormalities in standard X-rays.

KEY words: bull's head sign; chest pain; [^{99m}Tc]Tc-MDP scintigraphy

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Chest pain is a common reason for emergency department visits. If life-threatening cardiac and respiratory conditions are excluded, benign and more frequent conditions should be considered [1]. Non-cardiac chest pain (NCCP) is a wide group of disorders that could imitate acute coronary syndrome but with no evidence of heart disease in a conventional diagnostic process. In such a case, patients are often discharged with no certain diagnosis but rather with "diagnosis of exclusion" [2]. Because the condition is poorly understood, patients do not receive optimal care and pain can last for many years, causing recurrent visits in cardiac clinics, impairing quality of life, and leading to unnecessary cardiac medication [3].

We present a patient with chest pain related to rare spondyloarthropathy — SAPHO (synovitis, acne, pustulosis, hyperostosis, osteitis) syndrome. It is a rare condition (prevalence 1/10000) mainly affecting the anterior chest wall which is often misdiagnosed but could be detected by some characteristic features in scintigraphy imaging [4]. A 38-year-old female patient was admitted due to recurrent retrosternal chest pain lasting almost 5 years. A concomitant pain in the cervical spine and scapula was occurring periodically for several months. The tenderness of the anterior chest wall was observed. Past medical history included inflammatory low back pain lasting for many years, erythema nodosum, and presence of subchondral bone marrow edema in sacroiliac joints (in magnetic resonance). In the past she was treated for two weeks with diclofenac but because of dyspepsia, the treatment was discontinued. The patient was HLA-B27 positive. Inflammatory markers were increased C-reactive protein (CRP) = 18.4 mg/L and erythrocyte sedimentation rate (ESR) = 50 mm/h. Standard X-rays of the chest and spine were normal. Autoantibodies like rheumatoid factor or anti-citrullinated peptide antibodies were not detected. The occurrence of skin symptoms (erythema nodosum), presence of HLA-B27 antigen, and inflammatory low back pain suggest spondyloarthropathy as the reason of complaints. Due to sternoclavicular localization, and tenderness of the anterior chest wall, SAPHO

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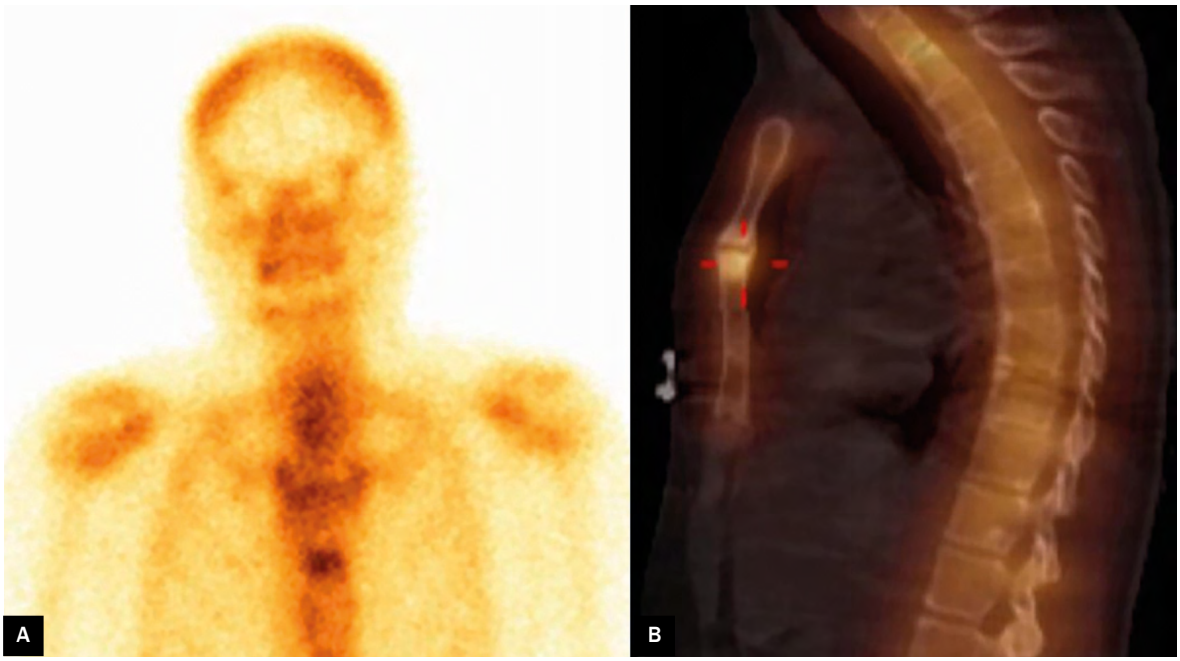

Figure 1. (**A**) Bone scintigraphy shows classic bull's head sign related to increased [^{99m}Tc]Tc-MDP, (**B**) sagittal image of SPECT-CT with increased osteoblastic activity in the sternoclavicular region

syndrome was taken into consideration. In further investigation, the patient underwent whole-body scintigraphy. The examination revealed increased radiotracer uptake in sternoclavicular, manubriosternal, sacroiliac joints, in the fifth rib and mandible. Increased osteoblastic activity (intense [^{99m}Tc]Tc-MDP) in manubriosternal and both sternoclavicular regions represents bull's head sign which is a rare finding, but pathognomonic to SAPHO syndrome (Fig. 1). SAPHO was diagnosed based on clinical sternoclavicular involvement and characteristic scintigraphy findings, despite the lack of skin lesions. Treatment with aceclofenac 100 mg twice daily with a proton-pump inhibitor was initiated. After 3-month therapy total remission was reached. SAPHO syndrome is rare or just unrecognized. Involving more advanced techniques of imaging like whole body scintigraphy shortens the time to make an accurate diagnosis. If we rule out this rare condition like SAPHO based on a lack of abnormalities in X-rays, the reason for the symptoms could be still unrecognized. Scintigraphy is valuable to show even subclinical areas of involvement, rule out malignancy, and infections, and to monitor treatment response [5]. Scintigraphy could help to avoid unnecessary hospital admissions and misguided therapy. The introduction of proper treatment improves substantially the quality of life.

Conflict of interest

There is no conflict of interest to declare.

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