

# SAPHO syndrome with mandibular manifestation

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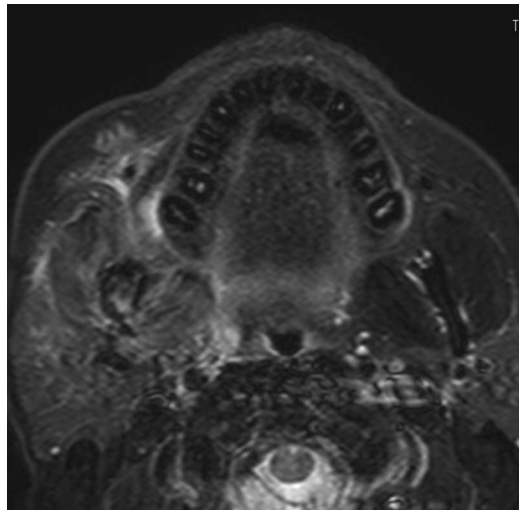
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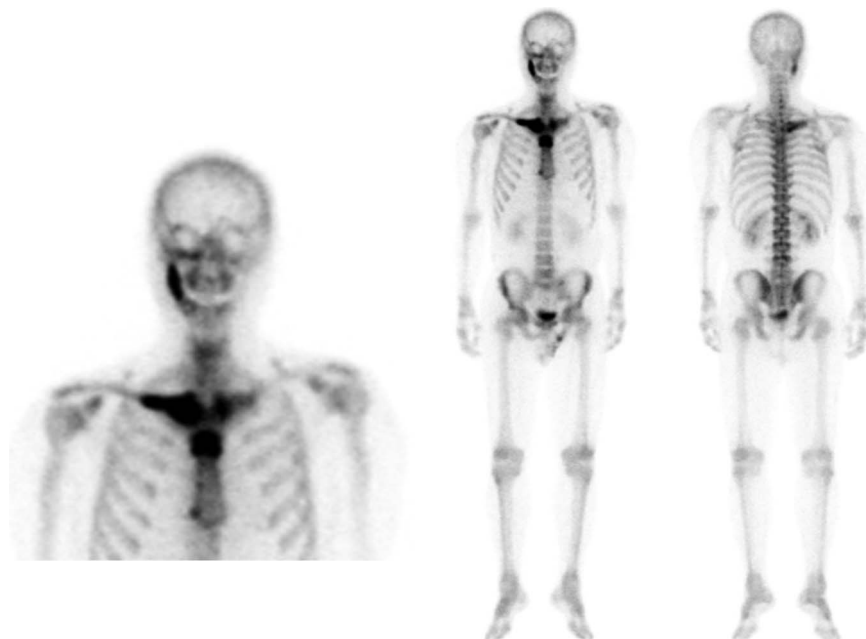
## DESCRIPTION

A 47-year-old man was referred to our hospital due to symptoms of trismus and exacerbation of palmo-plantar pustulosis. He had been presenting, for 20 years, with refractory and recurrent mandibular osteomyelitis accompanied by palmoplantar pustulosis and anterior chest pain—symptomatic treatment and surgery had been given at each



**Figure 1** MRI (short tau inversion recovery) showed high signal intensity in the right ramus of the mandible, with perilesional soft tissue swelling.

presentation. When the mandibular lesion got worse this time, he also had anterior chest pain. He had experienced similar episodes before this event. MRI (STIR: short tau inversion recovery) showed high signal intensity in the right ramus of the mandible, with perilesional soft tissue swelling (**figure 1**). These findings indicated active mandibular osteomyelitis. SAPHO syndrome was suggested by the clinical course and history. Bone scintigraphy using <sup>99m</sup>Tc-methylenediphosphonate (MDP) was performed and increased tracer uptake was seen in the right ramus of the mandible, greater wing of sphenoid bone, sternum and the sternocostoclavicular joints (**figure 2**). These characteristic clinical and radiological findings were compatible with the diagnosis of SAPHO syndrome involving the mandibular region. The term SAPHO syndrome was coined in 1987 to describe an association of characteristic bone, joint and skin lesions.<sup>1</sup> ‘SAPHO’ is an acronym for synovitis, acne, palmoplantar pustulosis, hyperostosis and osteitis. Its aetiology remains unknown.<sup>1 2</sup> The sternocostoclavicular, sternoclavicular, costoclavicular and manubriosternal joints are affected.<sup>3</sup> Bone scintigraphy using <sup>99m</sup>Tc-MDP is known to be important for diagnosis of this disease. SAPHO syndrome in the mandibular region is clinically rare and has been reported to be 10% of this entity.<sup>1</sup> SAPHO syndrome in the mandibular region is seen as recurrent mandibular osteomyelitis, mainly involving the body of the mandible.<sup>2</sup> In patients with clinical and radiological evidence of SAPHO syndrome, when recurrent mandibular



**Figure 2** Bone scintigraphy using <sup>99m</sup>Tc-methylenediphosphonate revealed increased tracer uptake in the right ramus of the mandible, greater wing of sphenoid bone, sternum and the sternocostoclavicular joints.



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osteomyelitis is also present, it should be considered as a part of SAPHO syndrome.

### Learning points

- ▶ SAPHO syndrome involving the mandible is clinically rare and has been reported to be 10% of this entity.
- ▶ SAPHO syndrome in the mandibular region is seen as recurrent mandibular osteomyelitis, mainly involving the body of the mandible.
- ▶ In patients with evidence of SAPHO syndrome, when recurrent mandibular osteomyelitis is also present, it should be considered as a part of SAPHO syndrome.

**Contributors** AB drafted the paper, and collected data and reviewed the text. All the authors diagnosed the disease, read and approved the final manuscript. ST and AK treated the patient and helped AB draft the paper. HO made the final revision.

**Competing interests** None declared.

**Patient consent** Obtained.

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