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Case report

Mandibular osteomyelitis related to SAPHO syndrome following dental implant surgery: A case report

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ABSTRACT

Chronic mandibular osteomyelitis is an intractable disease. There are some reports that it is related to the synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome. The etiology of the SAPHO syndrome is complex; it is likely to be the combined result of infectious, genetic, and immunologic factors. We present a case of mandibular osteomyelitis related to SAPHO syndrome that occurred after a dental implant surgery. A 56-year-old man presented at our hospital with the chief complaint of pain in the right lower jaw after dental implant treatment. We diagnosed bacterial osteomyelitis resulting from postoperative infection of the implant; his symptoms persisted despite antibacterial drug treatment and surgery. The patient also had a history of palmoplantar pustulosis and osteosclerosis of the collarbone. We diagnosed this patient with mandibular osteomyelitis related to the SAPHO syndrome. We initiated administration of 15 mg of prednisolone (PSL); however, the mandibular swelling could not be controlled with PSL alone. Thus, we initiated the administration of 5 mg of methotrexate and 15 mg of PSL, following which the cheek and jaw symptoms almost disappeared. The local bacterial infection and surgical stress in these patients with SAPHO syndrome may be related to the development of mandibular osteomyelitis. Thus, we do not recommend implant treatment for patients with the SAPHO syndrome.

1. Introduction

Mandibular osteomyelitis is one of the most common infectious diseases of the oral cavity and is usually odontogenic or traumatic in origin. Chronic mandibular osteomyelitis is associated with frequent remissions and exacerbations. The etiology of this type of osteomyelitis is still unknown. There are some reports that it is related to the synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome [1]. The SAPHO syndrome is a disorder characterized by pustular skin lesions and osteoarticular lesions and was discovered in 1987 by Chamot et al. [2]. SAPHO syndrome is a rare disease with a prevalence of 1/10,000 [3,4]. Although the diagnostic criteria for this syndrome have been proposed by some authors, the criteria given by Kahn, which was modified in 2003, seem to be the most precise [3]. Mandibular involvement in the SAPHO syndrome has been identified in about 10% of the cases [5]. There have been no reports on the trigger or cause of mandibular lesions in the patients with SAPHO syndrome. We report a case of mandibular osteomyelitis related to SAPHO syndrome that occurred after dental implant surgery.

2. Case report

The patient was a 56-year-old man who visited our hospital in February 2007 with the chief complaint of pain in the right lower jaw. He had received a dental implant in August 2006 at another clinic; however, the treatment had failed, and surgery for implant removal was performed a month later. The patient was diagnosed with palmoplantar pustulosis at the age of 42 years, and osteosclerosis of the left collarbone at age of 55 years. During the initial examination, we found swelling and pain on application of pressure in the right cheek, and desensitization of the mental region. Intraoral exploration showed a normal gingiva mucosa in the right retromolar area. Soluble

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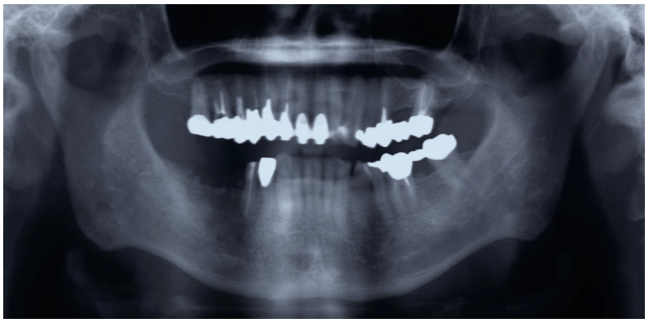

Fig. 1. Findings at the first visit in 2007. Panoramic radiograph showing soluble accentuation on the right side of the mandible. Mandibular osteomyelitis related to SAPHO syndrome following dental implant surgery: A case report.

accentuation was observed on the right side of the mandible on a panoramic X-ray image and computed tomography (CT) image (Figs. 1 and 2A). Scaly erythema with pustules was observed on the palm of the hands and soles of the feet (Fig. 3). The tonsils were not infected.

Initial diagnosis was bacterial osteomyelitis occurring after postoperative infection of the implant, and conservative treatment was attempted. Clarithromycin 400 mg/day and levofloxacin 300 mg/day was administered continuously. Moreover, a dermatologist started topical corticosteroids and vitamin D3 for the palmoplantar lesions. However, osteomyelitis was resistant to the antimicrobial treatment for 15 months. On CT image, we found soluble change mixed with sclerotic change and accompanied by bone enlargement on the right side of the mandible (Fig. 2B). According to bone scintigraphy, there was significantly increased accumulation on the right side of the mandible, left collarbone, chest bone, and both sternoclavicular joints (Fig. 4A).

In September 2008, the cortical bone was excised (decorticated)

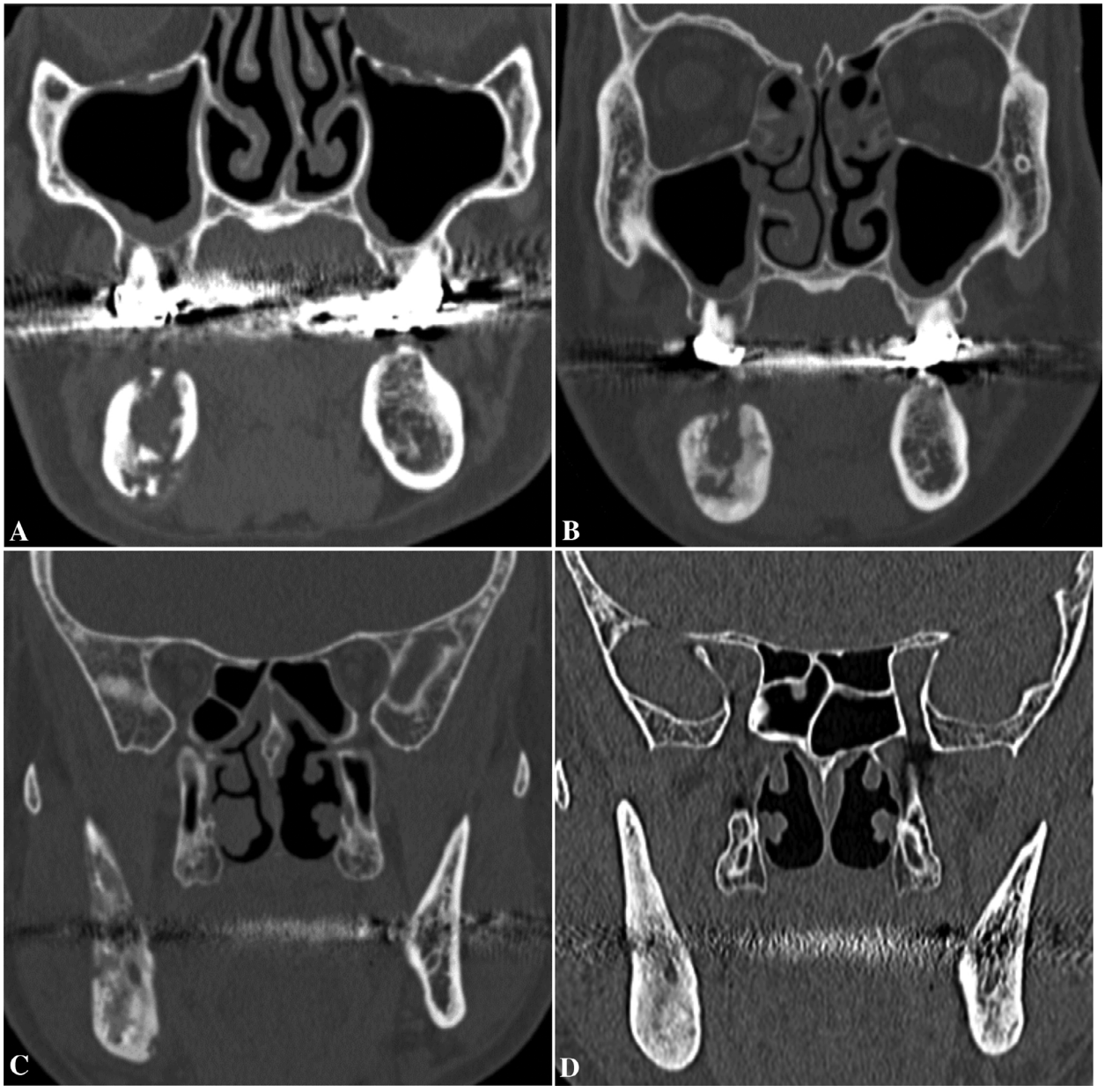

Fig. 2. Findings at the first visit in 2007. CT image showing soluble accentuation on the right side of the mandible. (B) Findings in May 2008. Change in the solubility and sclerotic changes were seen on the right side of the mandible and were accompanied by bone enlargement. (C) Findings after decortication in February 2010. Solubility and sclerotic changes have spread from the right mandibular ramus to the mental region. (D) Findings in 2015. Sclerotic change was observed in the entire mandible.