

原文題目(出處)：	Diffuse chronic sclerosing osteomyelitis of the mandible with synovitis, acne, pustulosis, hyperostosis, and osteitis: Report of a long-term follow-up case. J Oral Maxillofac Surg 2010;68:212-7
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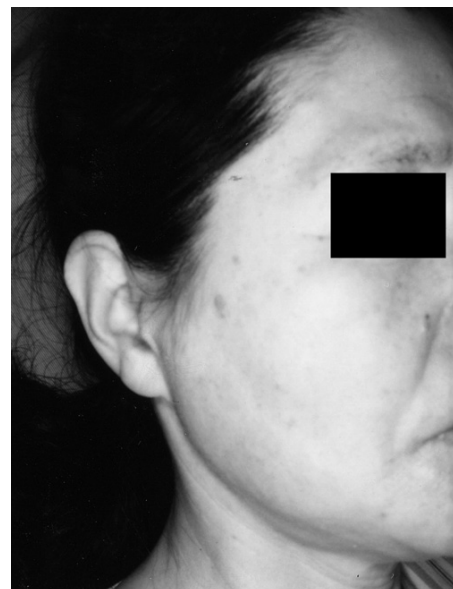
內文：

Introduction

- ✧ Mandibular osteomyelitis
 - ✓ one of the most common infectious diseases and usually odontogenic or traumatic in origin.
 - ✓ In 1987, Chamot et al described a SAPHO syndrome which is characterized by osteoarticular and dermatologic symptoms.
 - Synovitis,
 - Acne,
 - Pustulosis,
 - Hyperostosis,
 - Osteitis
 - ✓ Bone lesions in SAPHO syndrome demonstrate clinical and radiologic features similar to diffuse sclerosing osteomyelitis.
- ✧ SAPHO syndrome
 - ✓ The most prevalent site of bone lesions is the anterior chest wall (sternum, clavicles, ribs, spine, and peripheral long and flat bones)
 - ✓ Clinical diagnosis of SAPHO syndrome is defined as the presence of any one of the following:
 1. multifocal osteitis with or without skin manifestations
 2. sterile acute or chronic joint inflammation associated with pustules or psoriasis of palms and soles, or acne, or hidradenitis
 3. sterile osteitis in the presence of one of the skin manifestations
 - ✓ the etiology of SAPHO syndrome remains unknown.
 - ✓ Treatment has therefore been difficult and focuses on symptoms only.

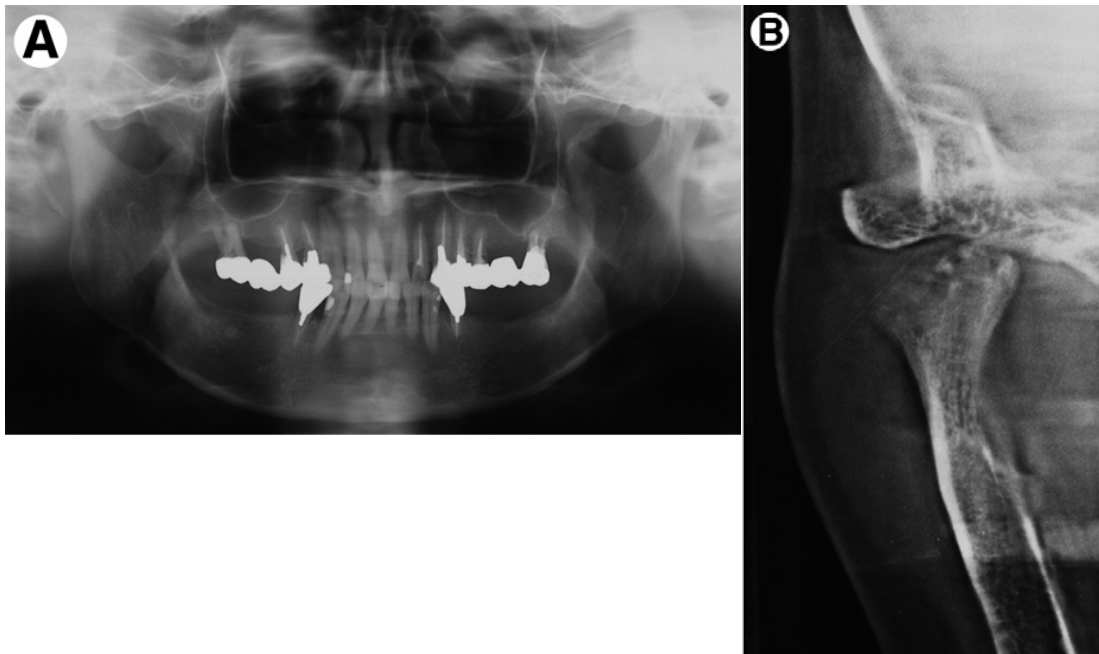
Report of a Case (long-term follow-up)

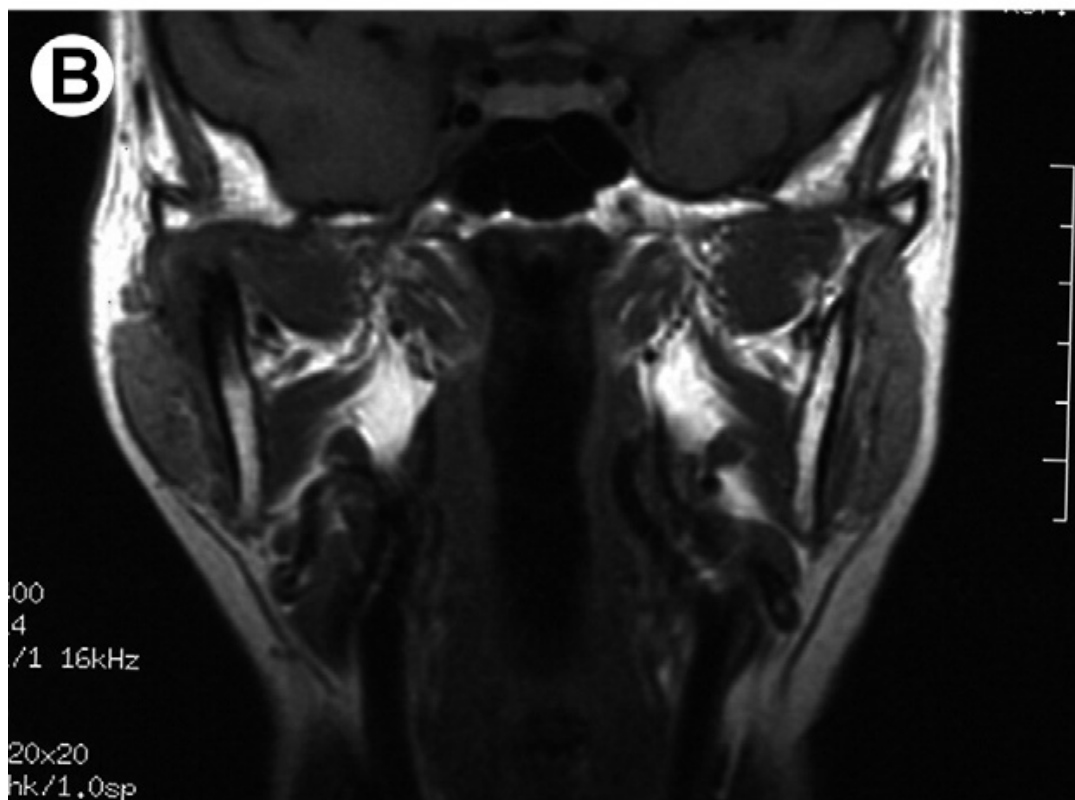
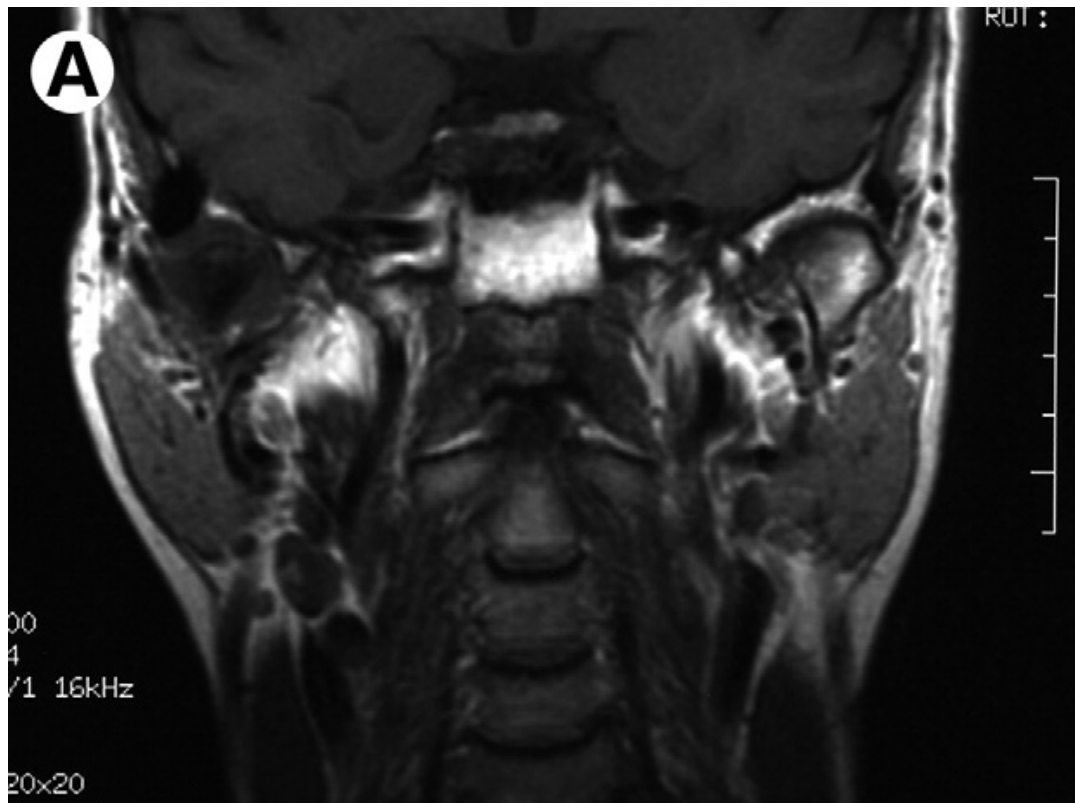
- ✧ General Data : A 51-year-old woman
- ✧ C.C. : a painful swelling of the right cheek associated with limited mouth opening for at least 3 weeks.
 - ✓ She did not have weakness or fever & reported no use of medications or previous treatment for these conditions.
- ✧ Medical history : no history of trauma to the maxillofacial complex.
- ✧ Clinical examination :



a bony hard swelling in the region of the right parotid-masseter and the right temporomandibular joint (TMJ) without suppuration and cervical lymphadenopathy.

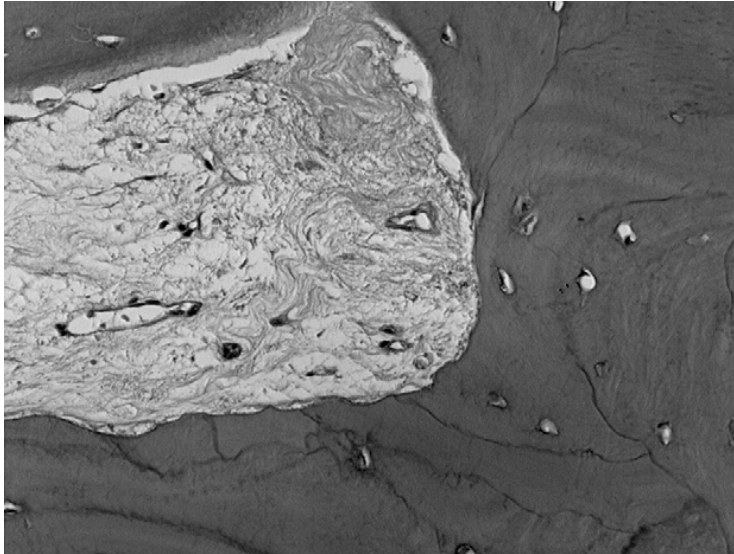
- ✧ Intraoral : no alterations were seen in the oral mucosa.
- ✧ In laboratory data : C-reactive protein was slightly elevated (2.3 mg/dL); other laboratory tests were within normal limits
- ✧ Panoramic radiogram (A) & coronal TMJ tomogram (B) :
destruction of the condyle and reactive sclerosis of the articular process of the mandible.





- ✧ Coronal contrast-enhanced T1-weighted magnetic resonance image: a low intensity signal of the bone marrow of the condyle (A) and ascending ramus (B).
- ✧ Diagnosis : mandibular osteomyelitis
- ✧ Treatment : antibiotic and NSAID therapy

- ✓ after 1 week, failed to improve the symptoms
- Partial resection of the condyle with an open biopsy
- ✧ Histopathologic examination :
 - fibrous granulation tissue and mature lamellate cellular bone

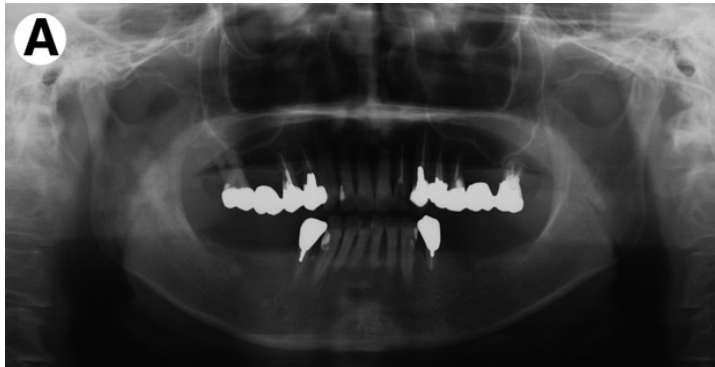


- ✧ Microbiologic culture from the biopsy specimen was

negative.

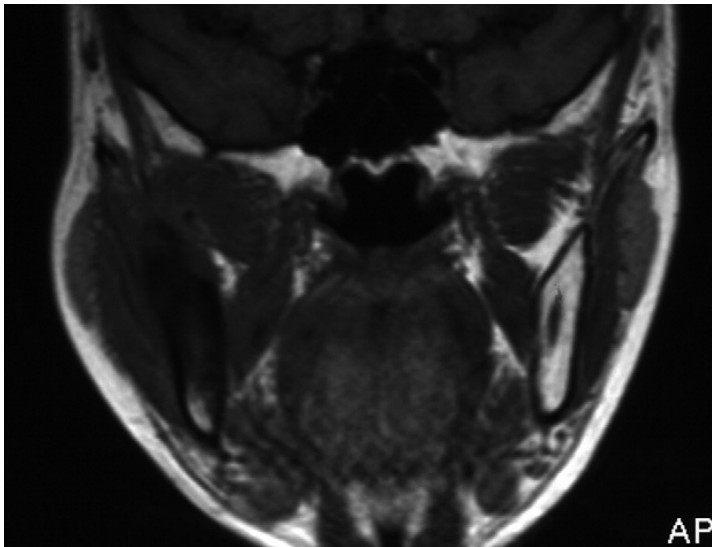
< two years later >

- ✧ Follow-up: experienced pain and swelling in the right mandibular body, but no evidence of recurrence of the mouth-opening limitation



- ✓ Panoramic radiogram :
 - bone sclerosis with scattered osteolyses of the ascending ramus.
- Coronal TMJ tomogram :
 - cortex formation of the condyle
 - progressive sclerotic change & periosteal reaction predominant in the ascending ramus
- Coronal contrast-enhanced T1-weighted MRI :
 - low intensity signal of the bone marrow to the mandibular angle,

- periosteal reaction observed outside the original cortex

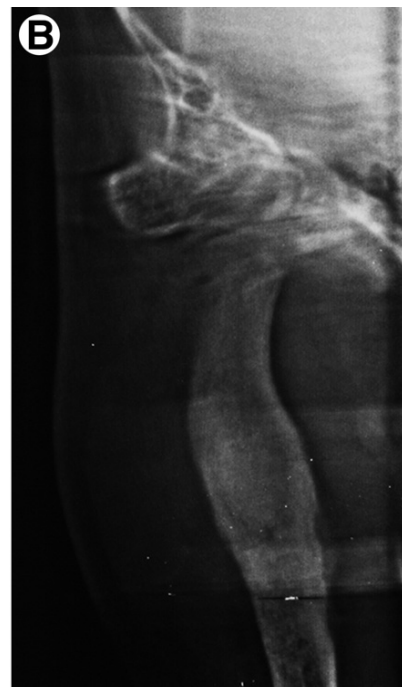
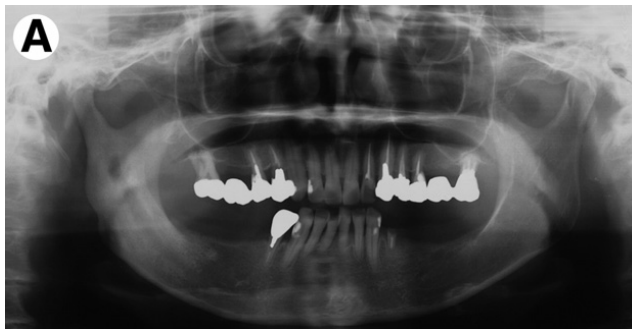


- Technetium 99m bone

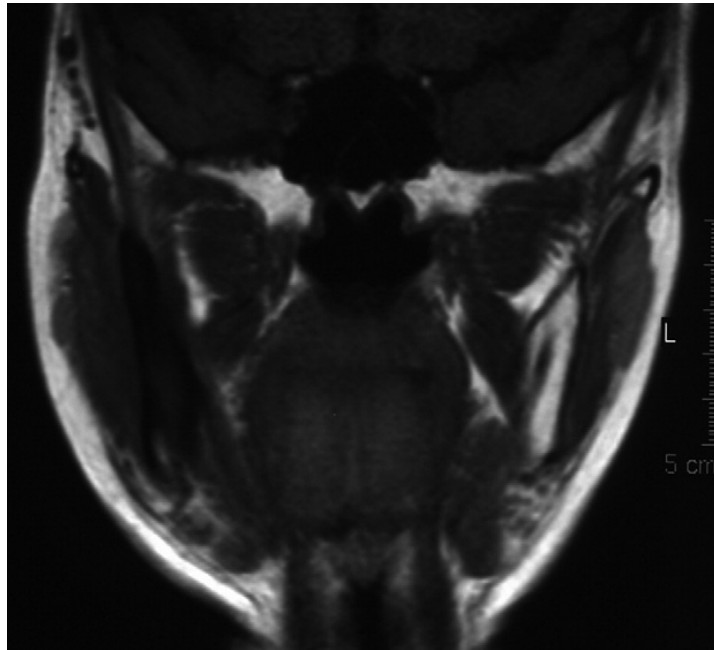
scintigram :

- enhance uptake in the right ascending ramus & sternoclavicular joint.
- ✧ Extensive decortication of the ramus was performed to decrease swelling of the mandible.
- ✧ Histopathologic findings : fibrous granulation tissue and bone fragments
- ✧ Microbiologic culture (-)
- ✧ clarithromycin(400 mg/d) and etodolac (200 mg/d) for at least 3 months.

< eight years later >



- ✓ Panoramic radiogram :
 - enhancement of sclerosis of the ascending ramus
 - Coronal TMJ tomogram :
 - enlargement of the ascending ramus
 - Coronal contrast-enhanced T1-weighted MRI : unaltered



Discussion

1. pediatric subset of SAPHO syndrome is

referred to as chronic recurrent multifocal osteomyelitis.

2. Diffuse sclerosing osteomyelitis of the mandible is a well-known bone lesion of SAPHO syndrome. (the most frequent is sternocostoclavicular lesion, followed by the sacroiliac joint and the spine)
 3. Various complication :
TMJ ankylosis ,
Inflammatory spread to the temporal bone causing deafness
 4. Swei et al recommended that mandibular osteomyelitis lesions should be classified into bacterial osteomyelitis and osteomyelitis in SAPHO syndrome
 - bacterial osteomyelitis : suppuration & osteolytic radiographic change with lamellar-type periosteal reaction
 - osteomyelitis in SAPHO syndrome : nonsuppurative and a mixed radiographic pattern accompanied by solid-type periosteal reaction, external bone resorption, and bone enlargement
 5. SAPHO syndrome :
 - Skin lesions typically seen in SAPHO syndrome are palmoplantar pustulosis and acne (84%)
 - It shows various immunogenetic backgrounds(antigen HLA B27)
 6. Up to now, treatment for SAPHO syndrome has focused only on symptoms
 - Surgical treatment (early stages), often has no or only short-term success
 - NSAIDs in combination with antibiotics (first choice)
 - Corticosteroid therapy (poor clinical response to NSAIDs)
- ✚ Recently, treatment with pamidronates or bisphosphonates has been reported to be effective, but no long-term data

Conclusion

- ✧ Mandibular osteomyelitis in SAPHO syndrome
 - ✓ nonsuppurative
 - ✓ mixed radiographic pattern accompanied by solidtype periosteal reaction
 - ✓ external bone resorption,
 - ✓ bone enlargement

題號	題目
1	<p>關於SAPHO syndrome,以下敘述何者不正確?</p> <p>(A) Surgical decortications has decreased the intensity and frequency of symptoms but has failed to resolve the process totally.</p> <p>(B) Although not found consistently,an increased prevalence of HLA 27 in patients with SAPHO has been noted by several investigators.</p> <p>(C) The cause is unkown.</p> <p>(D) Histopathologic studies reveal signs of infection rather than active bone remodeling.</p>
答案()	出處：Oral & Maxillofacial Pathology p.129 & p.130
題號	題目
2	<p>『SAPHO syndrome 在臨床上的表徵包括synovitis, <u>a</u>cne, <u>p</u>ustulosis, <u>h</u>yperostosis,和 <u>o</u>dontoma，這也是命名為SAPHO syndrome的由來。』請問以上這句話所描述的特徵，哪一點不是SAPHO syndrome的臨床表徵？</p>
	<p>(A) <u>s</u>ynovitis</p> <p>(B) <u>a</u>cne</p> <p>(C) <u>p</u>ustulosis</p> <p>(D) <u>h</u>yperostosis</p> <p>(E)<u>o</u>dontoma</p>
答案()	出處：Oral & Maxillofacial Pathology p.129