Spontaneous Regeneration of the Mandibular Bone Following Marginal Resection against Chronic Diffuse Sclerosing Osteomyelitis — Case Report —

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Accepted for publication on December 26, 1998

ABSTRACT. A 41-year-old Japanese man was referred to the clinic of the Department of Oral Surgery of Kawasaki Medical School Hospital, with a chief complaint of painful swelling of the right mandible. A biopsy specimen was suggestive of chronic osteomyelitis. A marginal resection of the inferior border of the mandible was performed and the remaining strut of the mandible was supported with a titanium implant plate. Histological examination of the resected mandibular bone was consistent with chronic diffuse sclerosing osteomyelitis. Healing of the wound was uncomplicated, but radiographic examination seven months later suddenly demonstrated a pathological fracture line without subjective symptoms. Three years later the pathological fracture line disappeared and a partial spontaneous regeneration of the mandibular bone was noted. The mainly spontaneous regeneration seems to be related to rigid fixation and periosteal capacity.

Key words: chronic diffuse sclerosing osteomyelitis — spontaneous regeneration — periosteal capacity

Chronic diffuse sclerosing osteomyelitis (CDSO), which mainly affects a large part of the mandible, is an uncommon disease characterized by repeated episodes of pain, swelling, and trismus. Because of an insufficient understanding of its etiology, different methods of treatment, including antibiotics, decortication, and hyperbaric oxygen, have been used. They have been helpful, but many patients continue to have relapses.[1,2] We experienced a rare case of partial spontaneous regeneration of the mandibular bone following a marginal mandibulectomy for CDSO, which relieved subjective symptoms. This report describes the clinical course of this case.

CASE REPORT

A 41-year-old Japanese man was referred to the clinic of the Department of Oral Surgery of Kawasaki Medical School Hospital on September 17, 1993, with a chief complaint of painful swelling of the right mandible of two months duration. He had received antibiotic therapy several times for the painful swelling, trismus and paresthesia of the lower lip before his first visit to our clinic. Maxillofacial examination disclosed expansion of the right mandible without trismus or paresthesia of the lower lip. Oral examination showed no

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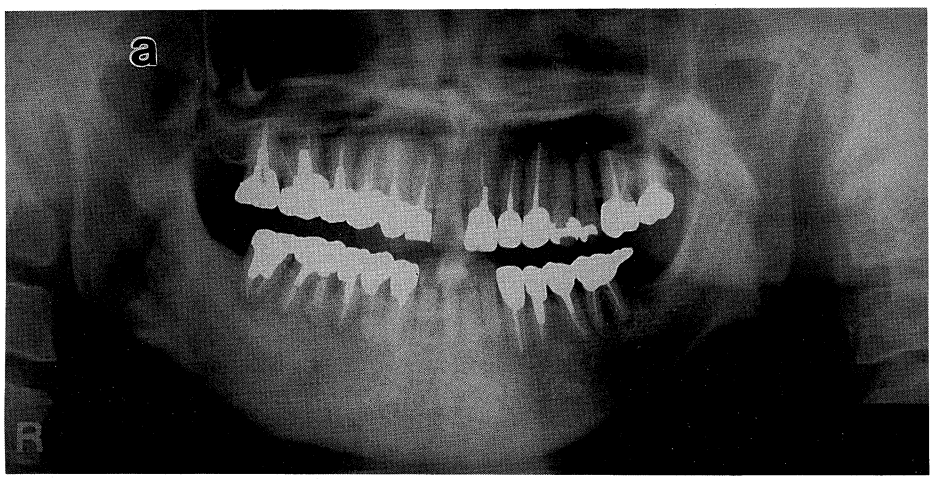
remarkable changes. A panoramic radiograph (Fig 1a) and an axial CT of the mandible revealed a ground-glass like osteosclerotic change in the right mandible, a finding of expansion of its inferior border and a thin radiolucent area around the root of the right mandibular second molar. Radioactive ^{99m}Tc-HMDP bone scintigraphy (Fig 1b) disclosed an increase in isotope



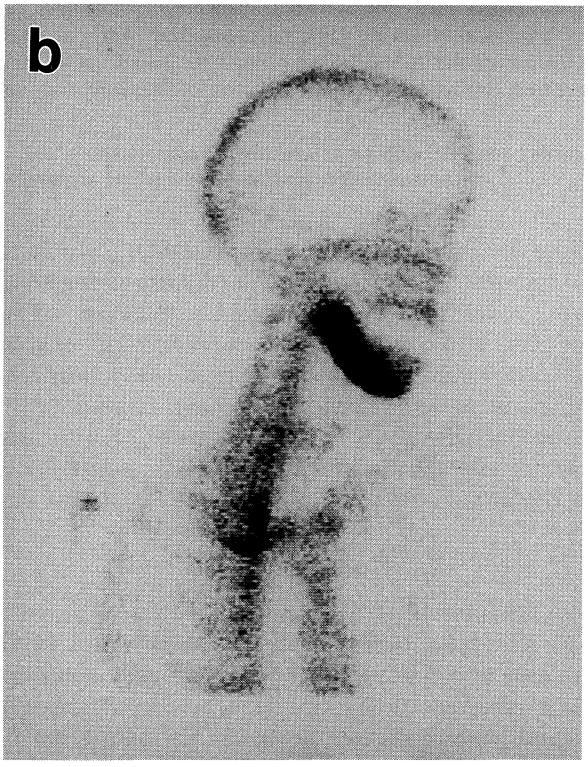


Fig 1. Radiographic examinations at the first visit
a. A panoramic radiograph
b. A bone scintigram of the mandible

uptake without any cold lesion in the right mandibular lesion. Radioactive ^{67}Ga citrate scintigraphy showed abnormal radioactivity in a similar geographic distribution. The uptake was consistent with the osteosclerosing findings on radiographs. The clinical diagnosis was chronic osteomyelitis or fibrous dysplasia with infection.

A biopsy specimen of the right mandibular bone was suggestive of chronic osteomyelitis. Under endotracheal anesthesia a marginal resection of the inferior border of the mandible from the right first premolar to the right mandibular angle was performed via a subperiosteal dissection. Since the remaining strut of the mandible was thin after resection, support was achieved with a titanium implant plate (Leibinger, Germany, Fig 2a). Healing of the wound was uncomplicated. Histological examination of the resected mandibular bone showed irregular bone trabeculae with a mosaic pattern. The bone trabeculae were accompanied by both osteoblastic rims and osteoclasts. The soft tissue between individual trabecula was fibrous and showed fibroblastic proliferation and lymphoplasmacytic infiltration. There was no evidence of neutrophilic infiltration, bacterial colonies or thrombophlebitis. These findings were consistent with chronic diffuse sclerosing osteomyelitis (Fig 3).

The patient was free of painful swelling, trismus and malocclusion. Radiographic examination seven months later suddenly demonstrated a pathological fracture line from the root apex of the right mandibular second molar to the upper margin of the resected lesion without subjective symptoms (Fig 2b, arrow). Thirteen months later this pathological fracture line had not changed. Three years later the pathological fracture line had disappeared and a partial spontaneous regeneration of the mandibular bone from the distal stump was noted radiographically (Fig 2c, arrowheads).

DISCUSSION

Decortication, which improves the circulation by establishing contact between the periosteal vessels and the subcortical tissues, is an acceptable treatment for CDSO, but relapses have occurred frequently after operation.[1,2] Therefore, in some cases, partial segmantal resection of the mandible has been employed : Nevertheless, all of these cases experienced recurrence within one year after removal of almost the entire radiographically and surgically affected area.[3] Recently van Merkesteyn and coworkers[4] advocated muscle relaxation treatment on the hypothesis that CDSO of the mandible could be a reactive hyperplasia of the bone caused by chronic tendoperiostitis probably due to muscular overuse. The results of this treatment were superior to the aforementioned results of other treatments. In the present case, we employed a marginal segmental resection of the inferior border of the mandible not only to remove as much of the affected bone tissues as possible, but also to preserve the continuity of the mandible. After marginal resection, the patient was free of painful swelling, but a postoperative pathological fracture of the remaining mandible because of occulusal force was observed. In recently reported cases of pathological fracture of the mandible resulting from chronic suppurative osteomyelitis after sequestrectomy or decortication, pathological fractures healed uneventfully after six to eight weeks of intermaxillary fixation by the ostegenic

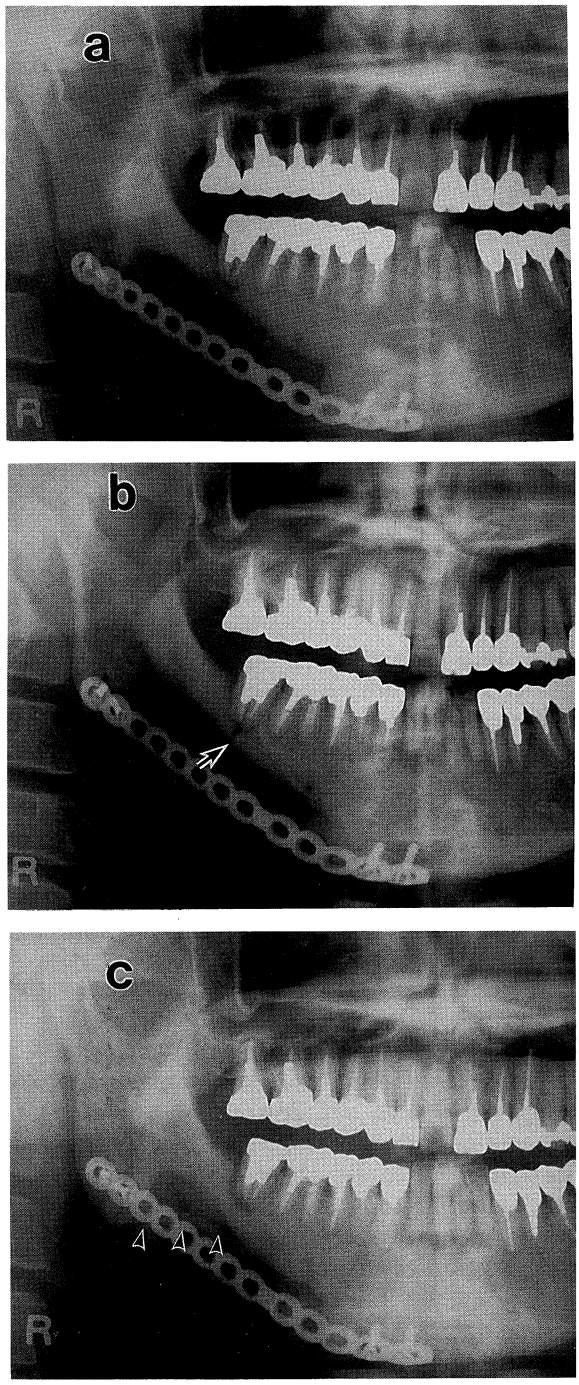


Fig 2. Panoramic radiographs after marginal mandibulectomy
a. Immediately after operation
b. Seven months later
c. Three years later, the pathological fracture line (arrow) has disappeared and partial spontaneous regeneration (arrowheads) of the mandibular bone was noted.

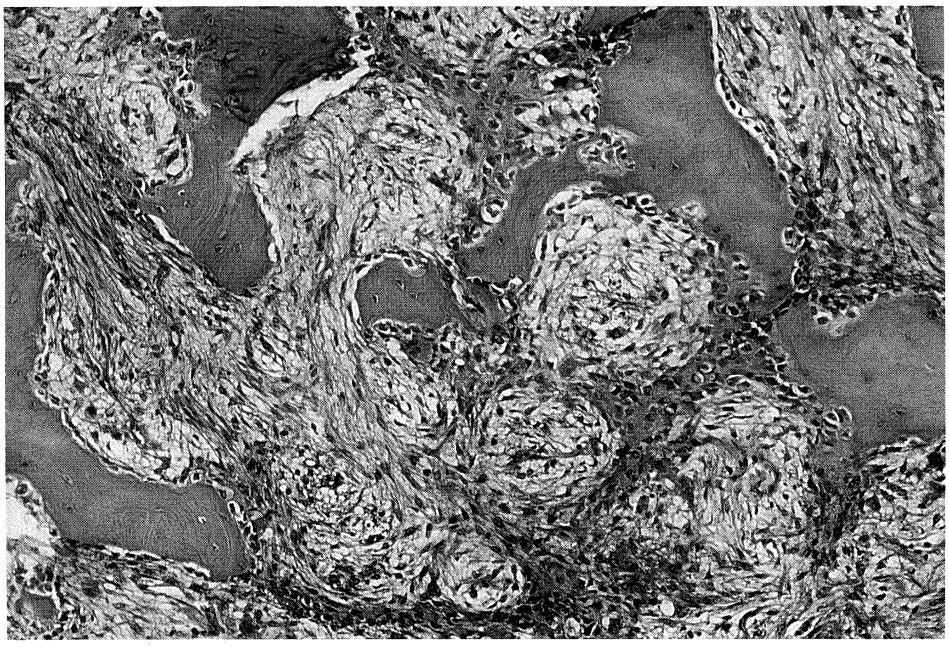


Fig 3. The resected mandibular bone shows irregular bone trabeculae and fibroblastic proliferation with chronic inflammatory infiltration (HE ×33).

potential of the remaining vital bone.[5] In cases of CDSO, although biopsy specimens have often shown remodeling of the buccal cortical plate with subperiosteal formation of new bone and a subcortical increase in trabecular bone volume,[2,4] healing of the pathological fracture has not been reported. In our case, the continuity of the remaining mandible was supported with a titanium implant plate and the pathological fracture remained asymptomatic. Therefore, we fully observed the condition of the fractured site for any change without medication.

Silbermann and coworkers[6] noted that osseous union occurred in the mandible in spite of an extensive bony defect resulting from sequestrectomy if osteomyelitis was controlled and adequate fixation was maintained. In the present case, most of the CDSO was resected by a marginal mandibulectomy and proper immobilization continued to be achieved with a titanium implant plate after the pathological fracture.

A rare case of spontaneous regeneration of the mandible following hemimandibulectomy has been reported in chronic suppurative osteomyelitis with a pathological fracture.[7] It was explained by the fact that the periosteum of the mandible regenerated the body of the mandible following mandibulectomy.[8] In our CDSO case, preservation of the periosteum, which restores the balance between available circulatory capacity and residual bone volume[1], may be a rational explanation for healing of the pathological fracture, and it also may have played a role in partial spontaneous regeneration of the mandibular bone.

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