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Oral Medicine/Case Report

A massive osteonecrosis with Le Fort I-type pathological fracture, caused by bisphosphonate-related osteonecrosis of the jaw (BRONJ): A rare case report



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ABSTRACT

Bisphosphonates (BPs) are useful drugs for the management of osteoporosis and metastatic bone disease, and are used to prevent skeletal-related events. However, the side effects of BPs, bisphosphonate-related osteonecrosis of the jaw (BRONJ), sometimes severely impairs patient quality of life. Here we report a case of severe BRONJ in the maxilla, which caused Le Fort I-type fracture-like bone fracture due to formation of a large sequestrum in the maxilla.

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1. Introduction

Bisphosphonates (BPs) are frequently prescribed for the treatment of patients with malignant bone diseases such as metastases from other solid malignancies [1] and multiple myeloma [2] and for the treatment of those with metabolic diseases such as osteoporosis [3]. However, as a complication of BP administration, BP-related osteonecrosis of the jaw (BRONJ) has been reported in a large number of patients in recent years. In the advanced stages, the presence of osteolysis extends to the jaw bone and forms sequestrum, resulting in bone weakness and pathological fracture [4]. The definition of pathological fractures is controversial, but the term can be described as fractures occurring as a result of normal function or minimal trauma in a bone weakened by pathological conditions

including metabolic bone diseases, severe infection, cysts and bone tumors. Diagnosis of the fracture itself is done based on physical and radiological examination. Generally, pathological fractures in the maxillofacial region occur in the mandible. To date, no advanced cases of pathological fractures in the maxilla caused by BRONJ have been reported. Here we report a case of BRONJ that progressed to a Le Fort I-type pathological fracture. Here we report a rare case of massive osteonecrosis caused by BRONJ with Le Fort I-type pathological fracture.

2. Case report

A 70s/M patient noticed swelling and discomfort in the right upper canine to premolar area along with a suppurative fistula, and he was referred to a general dental practitioner. At the clinic, he was treated by irrigation of the swollen area and a clinical diagnosis of dental infection was made. However, he experienced absolutely no improvement after the treatment. Two months later, he noticed greatly increased mobility in the upper canine; thus, the canine was extracted by dentist. During the following month, his bilateral first premolars, right second premolar, and first molar in the maxilla spontaneously fell out. A partial upper denture was subsequently created. Soon afterward, the patient noticed swelling

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and pain of the left upper premolar to molar area and was again referred to a general dental practitioner. His left second premolar and first molar in the maxilla were found to be highly mobile; thus, they were extracted in a month later from denture creation. After that, bilateral second molars in the maxilla spontaneously fell out. Although the teeth extraction sockets failed to heal well, a complete upper denture could be remanufactured as there were no abnormalities of the oral mucosa, including the palate. However, several weeks later, the denture became unstable, so it was lined using a soft denture-lining material. After the denture was adjusted, it had to continuously remain in his oral cavity for few days. At the end of this period, he was referred to a dental clinic for removal of the denture lining, which proved to be difficult. When attempting to remove the denture, the dentist noticed shaking of the maxilla along with the denture. The patient was then referred to our service of Oral and Maxillofacial Surgery Division, Kagawa Prefectural Central Hospital for further evaluation and treatment of the whole maxillary movement.

His medical history included prostate cancer. He had suffered from pain due to bone metastasis, and he had been treated with monthly therapy with zoledronate (4 mg), which was intravenously administered, for approximately 27 months. However, he and his family did not mention this to the dentist as they believed that there would be no relationship between his dental problems and the use of BPs for past several years.

Extraoral examination detected a cutaneous discharging fistula in the right side wall of the nose area (Fig. 1A). The fistula was in communication with the corresponding right apical maxillary canine region. He had hypoesthesia of the bilateral infraorbital nerve distributions. Symptomatic maxillary sinusitis was not observed. His maxilla was mobile with no simultaneous movement of the nasal bones upon palpation. He was unable to bite by maxillary movement, and the denture was unsuitable. Intraoral examination revealed exposed necrotic bone coupled to his denture at the site of total maxilla (Fig. 1B).

Computed tomography (CT) revealed fractures of the left pterygoid plate, bilateral anterior and lateral maxillary walls, and posterior nasal septum in the axial bony window (Fig. 2A). Coronal views revealed bilateral fractures through the lateral walls of the maxillary sinuses (Fig. 2B). A three-dimensional reconstruction view allowed clear visualization of the fracture line at the Le Fort I level (Fig. 2C). Although no lesions were observed in the skull base, evidence of inflammation was observed from the right part of the sphenoid to the external surface of the skull base.

Under local anesthesia, sequestrectomy of the bilateral total maxillary bone along with his denture was performed. The maxilla was easily removed as it was attached to a region of palatine mucous membrane only. The necrotic bone with irregular ragged margins of the cortex, was removed *en block* with the maxillary denture firmly attached to the bone (Fig. 3). After removal of the maxillary sequestrum, the discharging right cutaneous facial fistula gradually decrease, his fistula disappeared in a month after surgery.

Histopathological evaluation of the resected maxilla confirmed necrosis of the maxillary bone with bacterial overgrowth and no evidence of metastatic disease.

On the basis of clinical, histological, and instrumental examinations, osteonecrosis of the jaw induced by the previous treatment with zoledronate was diagnosed.

3. Discussion

BRONJ was first reported as a serious side effect of long-term BP treatment by Marx [5] in the United States in 2003. The mandible is more commonly affected than the maxilla [6–8]. The exact reason for the characteristic distribution of osteonecrosis in the jaws is unknown; several reasons have been postulated such as the comprehensive blood supply, different bone architecture, and different embryological background [4,9]. This case demonstrated that BRONJ can affect maxillary bone. In addition, this was a rare case of osteonecrosis affecting such a large sequestrum in the maxilla.

BRONJ mainly occurs after dentoalveolar surgery, including procedures such as tooth extraction, periodontal surgery, and dental implant placement [6,7]. BRONJ may also occur spontaneously without any apparent dental disease, treatment, or trauma. In this case, spontaneous loss of bilateral premolar and molar teeth in the maxilla had previously occurred. Therefore, etiology of spontaneous BRONJ was suspected. When the maxillary denture was fitted, this may have caused the sequestrum to spread to the entire maxilla.

After performing denture adjustment, the sequestrum had become strongly adhered to the denture through the oral mucosa and denture lining material. As a result of the weak downward force exerted on the molars around the anterior maxillary portion to remove the denture, a pathological fracture resembling a Le Fort I-type fracture was caused. Maxillary fractures most frequently occur as a result of high energy trauma caused by assaults, sport injuries, or traffic accidents. In contrast, in this case,

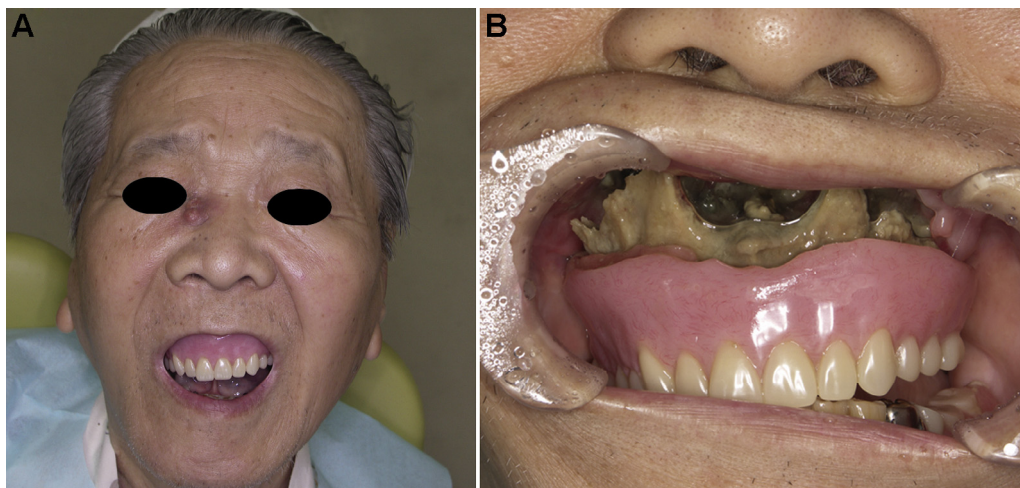


Fig. 1. (A) The patient was unable to close his mouth by maxillary movement, and the denture was unsuitable. (B) Intraoral examination revealed exposed necrotic bone connected to his denture at the site of total maxilla.

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