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Pathological mandibular fractures following idiopathic resorption of bilateral mandibular rami and coronoid processes

KEYWORDS
Coronoid process;
Idiopathic mandibular resorption;
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Pathological fracture

Pathological mandibular fractures are rare, accounting for fewer than 2% of all mandibular fractures.[1] Pathological fractures usually may follow surgical interventions such as third molar removal or implant placement, and are caused by osteomyelitis, osteonecrosis, cyst, or tumor.[1] Therefore, idiopathic mandibular fracture is rarer, especially the bilateral fractures. Here, we reported a rare case of pathological mandibular fractures following idiopathic resorption of bilateral mandibular rami and coronoid processes.

A 74-year-old man with open bite and bilateral mandibular pain and swelling was referred to our department. Panoramic radiograph showed bilateral mandibular fractures caused by resorption of the bilateral mandibular rami and coronoid processes (Fig. 1A). Panoramic radiograph taken 20 months before the initial visit revealed resorption of the bilateral mandibular angles (Fig. 1B). Three-dimensional computed tomography (3D-CT) showed open bite and bilateral mandibular fractures (Fig. 1C and D). Bimaxillary removable partial prostheses were fabricated to improve occlusion. Although several examinations were performed to identify the cause of mandibular resorption, systemic sclerosis with oral manifestations of malocclusion and mandibular resorption was ruled out. Panoramic radiograph taken 2 months after the initial visit revealed further resorption of the bilateral mandibular fracture sites (Fig. 1E). However, there was slight resorption of the mandibular fracture sites in 3D-CT at 6 months after the first visit (Fig. 1F and G). Therefore, mandibular reconstructive surgery will be performed, when mandibular resorption does not occur after the follow-up of more than 1 year from the first visit.

Progressive osteolysis or resorption of the mandible is a rare entity. Mandibular resorption in systemic sclerosis predominantly occurs at areas of muscle attachment, such as the mandibular angle, coronoid process, and mandibular condyle.[2] The current theory behind the etiology of bone resorption is multifactorial, including ischemia secondary to pressure necrosis from fibrosis and atrophic musculature, as well as decreased vascularity as a result of vessel wall sclerosis.[2] Gorham's disease (GD) is an extremely rare bone disorder characterized by osteolytic bony resorption and can affect any bone in the body.[3] It is of unknown etiology and thought to result from localized endothelial proliferation of lymphatic vessels that cause bony resorption.[3] The diagnosis can only be made after excluding other common causes of bony resorption such as those caused by infection, inflammation, malignancy, and endocrine.[3] In maxillofacial GD, the mandible is more commonly affected than the maxilla.[4] Maxillofacial GD can affect any age group, and most cases are detected before 40 years of age, with a mean presenting age of 29.6 years.[4] The mobility of teeth can be the initial presenting finding, and the jaw continues to resorb and eventually disappear completely.[4] The

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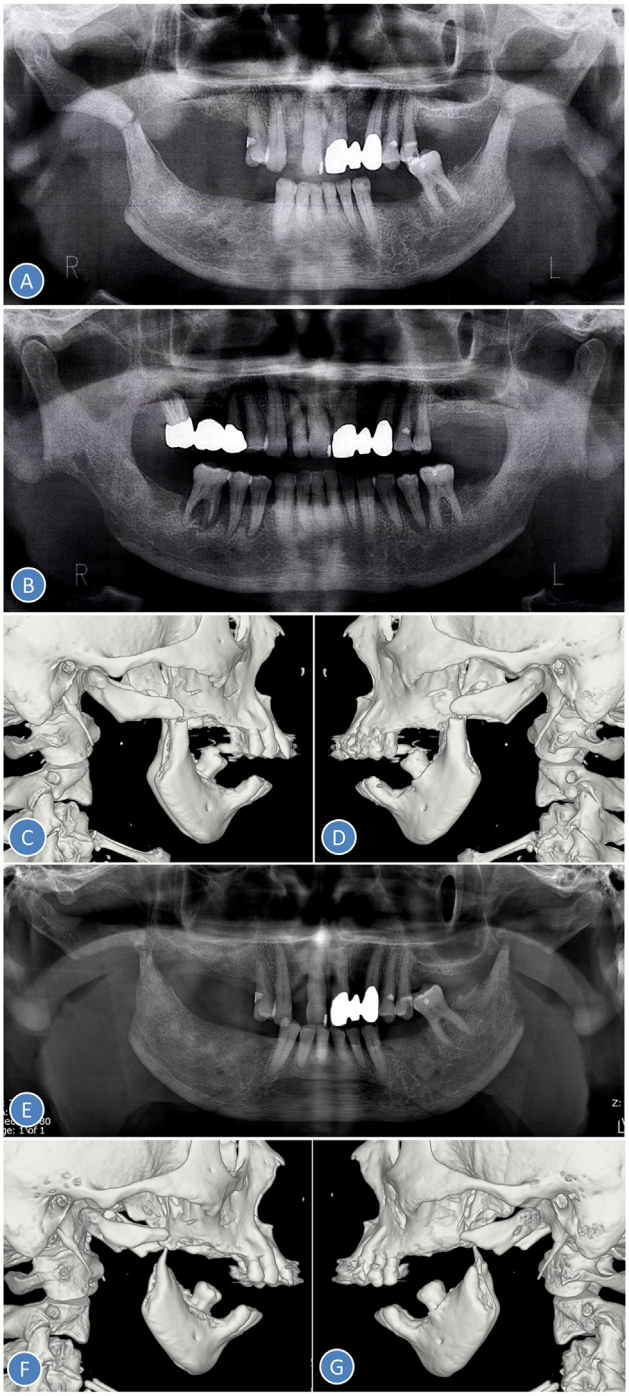

Figure 1 (A) Panoramic radiograph at the initial visit, (B) Panoramic radiograph at 20 months before the initial visit, (C, D) Three-dimensional computed tomography at the initial visit, (E) Panoramic radiograph at 2 months after the initial visit, (F, G) Three-dimensional computed tomography at 6 months after the initial visit.

differential diagnosis includes Paget disease of bone, Langerhans cell histiocytosis, osteomyelitis, primary intraosseous malignancy, metastatic malignancy, hyperparathyroidism, aneurysmal bone cyst, and rheumatoid arthritis.[4]

In the present case of an elder, spontaneous resorption of bilateral mandibular angles and coronoid processes resulted in a pathological mandibular fracture. However, the cause of the mandibular resorption was not clear, because the patient refused a biopsy for the diagnosis. Once bilateral mandibular resorption has stopped, we will plan mandibular reconstruction with the biopy.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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