

Delayed Presentation of Mandibular Osteonecrosis Following Herpes Zoster Infection - A Case Report

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Abstract

Rationale: Apart from the usual presentation of herpes zoster (HZ) infection (HZI), reports of spontaneous teeth exfoliation and osteonecrosis are infrequent and sporadic. **Patient Concerns:** A 51-year-old male patient presented with spontaneous exfoliation of multiple teeth and subsequent pathological fracture on the right side of the lower jaw after three months of HZI. **Diagnosis:** Biopsy was taken from the alveolar bone of the oedematous region, which revealed the presence of trabeculae of dead bone with empty lacunae. **Intervention:** Necrosed part of the alveolar bone was excised under local anaesthesia and antibiotic coverage, which was followed by open reduction and internal fixation of the pathological fracture under general anaesthesia. **Outcomes:** The patient was followed up for one year without any evidence of recurrences. **Take-away Lessons:** Presentation of osteonecrosis following HZI is unique but rare and should be diagnosed at the earliest.

Keywords: Fractures, herpes zoster, osteonecrosis, spontaneous, tooth exfoliation

INTRODUCTION

Osteonecrosis of the jaw is a term applied to a condition characterised by the persistence of symptoms such as delayed healing of extraction socket in the mouth, exposed bone in the oral cavity, development of sequestra or presence of communication in between oral cavity and skin or maxillary antrum.^[1] It directly harms bone cells and soft tissue from high-potency bisphosphonates or other cytotoxic drugs such as methotrexate, etanercept, adalimumab and rituximab.^[1,2] Other conditions which precipitate this condition include myeloma, malignancies of other organs such as the breast and prostate, ischaemia of bone, disease-causing low bone turnover, persistent bony infection and osteoporosis.^[1,2] A significant correlation has been found between trigger events and medication-related osteonecrosis of the jaw (MRONJ) in osteoporotic patients in recent studies, concluding the role of the trigger factor.^[2] Documented presentations of osteonecrosis after herpes zoster (HZ) infection (HZI) are rare.^[3,4] Unilateral involvement of jaw bone with spontaneous exfoliation of teeth is typical of the condition. This case report describes such a rare presentation of osteonecrosis of the mandible of post-herpetic aetiology.

CASE REPORT

A 51-year-old man was referred by his attending dentist to our Institute's Department of Oral and Maxillofacial Surgery with the chief complaint of spontaneous exfoliation of multiple teeth from the right lower jaw region. The patient had a history of fever with unilateral skin rashes three months back with numbness from the right angle of the mouth to the area in front of the right ear, followed by the appearance of vesicles. The unilateral distribution of scars of healed vesicles was noted on the day of the patient's first visit to the department [Figure 1a]. The fever lasted for seven days and remitted on its own. It was diagnosed by his attending physician as an HZI and was treated with oral paracetamol without any antiviral therapy. One month later, during the convalescent period, spontaneous mobility of teeth

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was noticed on the right lower back jaw region. The mobile teeth exfoliated subsequently with the progressive mobility of teeth in the anterior region. Intraoral examination revealed an edentulous region in the previously exfoliated teeth region, covered with normal gingival mucosa [Figure 1b], without showing any necrotic alveolar bone, discharge or tenderness. The teeth in the anterior region had Grade II mobility without any tenderness on percussion. Cone-beam computed tomography (CBCT) revealed rarefaction of the alveolar bone of the right mandible [Figure 2], with an appearance of mixed osteolysis, and dense radiographic areas confined within the alveolar bone without extending to buccal or lingual cortical plate (which was evident in the axial and coronal views of CBCT images). The necrotic zone was poorly defined in the periphery, with no periosteal apposition and no border that could suggest sequestration. The patient did not have any history of underlying medical conditions which would affect his immunity as a whole. The case was provisionally diagnosed as right mandibular osteonecrosis following HZI in light of the medical history, topographical arguments and literature reviews.

A biopsy was planned from the edentulous region to confirm the diagnosis. The mobile anterior teeth were removed, which had a poor prognosis, along with the removal of a sufficient amount of necrotic bone from the alveolus without much damaging the buccal cortical plate using bone nibblers and bone files under local anaesthesia [Figure 3]. The patient was kept on antibiotic

therapy (amoxicillin 1 g q8 h for seven days and metronidazole 500 mg q8 h for seven days). The bony tissues were sent for histopathological examination, which evoked the presence of trabeculae of dead bone with empty lacunae and surrounded by new bone formation with osteoblastic lining [Figure 4]. The overall histopathological features were suggestive of osteonecrosis of the right mandibular alveolus. One week later, post-operative control was satisfactory; the oral mucosa was healing, and the patient had no symptoms. On his regular clinical follow-up after two months of biopsy, a panoramic radiograph was suggested for the evaluation of the disease process, which revealed a pathological fracture of the right body of the mandible anterior to the angle region [Figure 5]. The possibilities of any iatrogenic causes for the spontaneous fracture of the weakened bone were ruled out. The fracture was treated with three weeks of intermaxillary fixation (IMF), followed by open reduction and internal fixation with a titanium reconstruction plate and screws under general anaesthesia. On follow-up after one week, the surgical wound revealed uneventful healing, and during the follow-up of one year, there were no signs of recurrences both clinically [Figure 6] and radiographically [Figure 7].

DISCUSSION

There are 1.5–3 cases of HZI/1000 subjects, which increases to 10/1000 in advanced ages.^[5] Rose *et al.* in 1908, followed



Figure 1: (a) Extraoral view showing the presence of scars of vesicles present unilaterally on the right side of face. (b) Intraoral view showing normal epithelium over the edentulous region of the right lower arch



Figure 2: Axial, coronal and panoramic view of cone-beam computed tomography showing rarefaction of alveolar bone on the edentulous region of right side of mandible

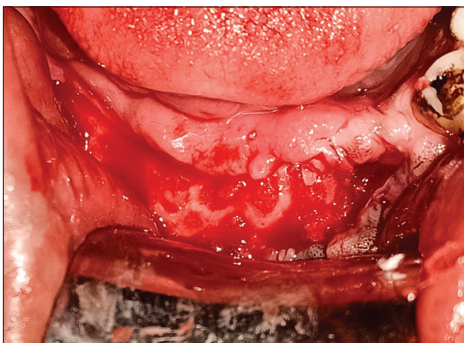


Figure 3: Intraoral view during the removal of necrotic bony tissue from alveolar bone

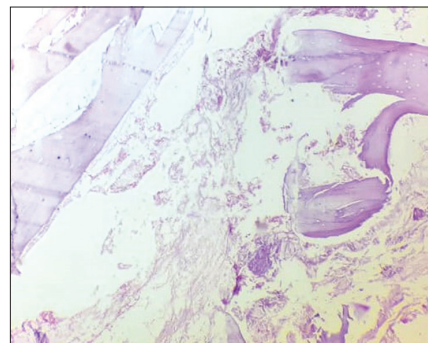


Figure 4: Histopathological features were suggestive of osteonecrosis, magnification value is 10x

by Gonnet *et al.* in 1922, described bony alterations following shingles with dental exfoliations following HZI.^[6] Initially, the occurrence of osteonecrosis after HZI was restricted to the mandible of elderly people, but later on, similar occurrences were found in the maxilla also.^[6] Wright *et al.* in 1983 and later Mendieta *et al.* in 2005 presented a review of cases with the comparison of clinical features associated with osteonecrosis following HZI.^[6,7] In their review, the age range was found to be between 6 and 85 years, with female preponderance. Ten of them did not have any underlying systemic disorder. The relationship between the occurrence of such osteonecrosis to medically compromised conditions was indecisive. In our report, the patient did not have any underlying systemic condition.^[7] There was no history of antiviral treatment during the acute course of the

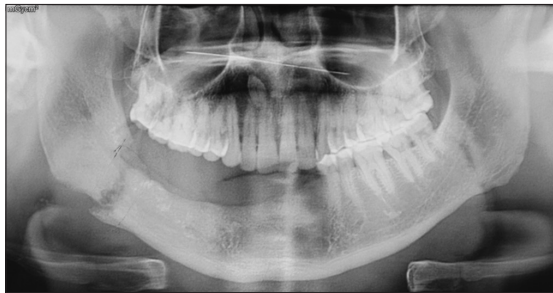


Figure 5: Panoramic radiograph showing pathological fracture of the right body of mandible during follow-up after removal of necrotic bony tissue



Figure 6: (a) Extraoral view during follow-up after one year showing reduced amount of residual facial scar. (b) Intraoral view during follow-up after one year showing no signs of recurrences



Figure 7: Cone-beam computed tomography showing signs of bony healing after one year of open reduction and internal fixation of the pathological fracture

disease in our case report, although most of the literature mentions antiviral therapy. Pre-existing pulpal or periodontal pathology, trauma to the jaw bone were suggested as contributory factors.^[7] In our report, the patient did not have either of them. The occurrence of bacterial superinfection is inconspicuous and difficult to diagnose and treat the root cause of the disease. In that case, salivary polymerase chain reaction aids in definitive diagnosis of the virus.^[8] Antiviral regimen was avoided in the report as the disease was less destructive in its presentation. The pathological fracture was detected on the routine follow-up after two months of the excision of necrotic bone from the alveolar region. It was asymptomatic and without any history of local causes, i.e. trauma. Loss of vertical bone height after the excision of necrotic bone may be contributory. Pichardo *et al.*, in their observational study, treated seven out of 15 patients with pathological fractures of the mandible in MRONJ with IMF of six to eight weeks.^[9] Out of seven patients, four patients developed pseudoarthroses, and the rest showed a good amount of bony healing. Pathological fractures after initial surgical intervention occurred due to a loss of vertical bone height.^[9,10] Conventional Champy plate fixation was inadequate due to atrophied bone, which had the chance for the development of plate exposure, wound dehiscence and plate infection.^[9,10] Keeping these possibilities in mind, the patient in our case report was treated with fixation at the lower border of the mandible with a titanium reconstruction plate which showed appreciable bony healing on follow-up.

CONCLUSION

Predisposing factors jeopardise the process of recovery from such type of osteonecrosis. Hence, more literary support is required for evidence-based treatment protocol.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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