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Spontaneous Tooth Exfoliation Secondary to Osteoradionecrosis of Jaw Bones - A Case Report

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ABSTRACT

Osteoradionecrosis of the jaws is a serious and grave sequalae of therapeutic radiotherapy given for head and neck malignancies. It is a delayed process where irradiated bone becomes necrosed leading to its exposure. The most common signs and symptoms are localized pain, dysesthesia, bone exposure, sino-cutaneous or oro-cutaneous fistula, trismus, and pathological fractures.
This article describes a documented instance of osteoradionecrosis affecting the jaws. The patient in this case experienced exposed and necrotic bone in both the anterior maxilla and mandible within six months after undergoing radiotherapy for grade III choroid plexus carcinoma.

Keywords: Radiotherapy, Osteoradionecrosis of jaw, Necrosis

INTRODUCTION

Osteoradionecrosis (ORN) is classically defined as "exposed bone through an opening in the overlying skin or mucosa, persisting as a non-healing wound for three months or more".[1] Changes in bone that occur following radiotherapy were first identified and reported by Ewing in 1926. He referred to this condition as radiation osteitis. The first documented case of ORN affecting the jaws following radiation therapy was published much later, specifically in 1992, by Regaurd.[2]

ORN is a delayed consequence of radiotherapy.[2] It usually manifests within 2 years if the patient has received a high dose of radiation therapy (>70 gray). On the other hand, late presentation is typically associated with trauma and impaired tissue healing.[3] Symptoms of ORN include pain, halitosis, dysgeusia (altered taste sensation), dysesthesia (abnormal sensations) or anaesthesia (loss of sensation), trismus (inability to fully open the mouth), difficulty in chewing and swallowing, speech difficulties, fistula formation (an abnormal connection between different body structures like tissues or organs), pathological fracture and localized spread of infection.[4]

The incidence of ORN in individuals with head and neck cancer is approximately 7.4% following conventional radiotherapy, 5.1% after intensity-modulated radiotherapy, and 6.8% after chemo-radiotherapy.[5]

This case report describes a patient who developed necrosis within six months of receiving radiation therapy for grade III choroid plexus carcinoma. The affected individual exhibited necrotic bone exposure in both the maxilla and mandible.

CASE REPORT

A 23-year-old male patient reported to the department of Oral Medicine and Radiology of Government Dental College and Hospital, Kadapa with the chief complaint of spontaneous exfoliation of teeth a month ago while doing mouthwash.

Exfoliation began with a solitary tooth, the right central incisor of the maxilla. This was followed by exfoliation of the lateral incisors and central incisors of the mandible. The patient reported experiencing difficulties in mouth opening and eating, as well as intolerance to spicy food. However, the patient did not report any notable changes in salivary flow and had no history of undergoing any extractions.

The patient's medical history revealed that he was diagnosed with grade III choroid plexus carcinoma with D1-D2 intradural extramedullary metastasis one year ago. He underwent a right fronto-temporo-parietal craniotomy and complete tumour removal, followed by craniospinal irradiation at a dose of 54.8Gy administered in 30 fractions, each delivering 1.8Gy per fraction. In addition to the surgery and radiation therapy, the patient also received one cycle of adjuvant chemotherapy of ICE regimen. This involved injections of Ifosfamide at a dosage of 5mg/m², Mesna at a dosage of 5g/m², Carboplatin with an AUC value set to 5, and Etoposide at a dosage of 100mg/m².

FIGURE 1 A & B – Patient's Right Profile Showing the Scar from the Craniotomy (Figure 1a) And Front Profile (Figure 1b)

During the general examination, the patient was alert and coherent. The patient had a moderate build with average height and exhibited a reduced range of mouth opening measuring approximately 28 mm.

Intraorally, exposed devitalized and necrotic bone through ulcerated mucosa with mild yellowish discoloration was seen in maxillary alveolus and mandibular central incisor region with empty sockets (Figure 2a and 2b). The alveolus was denuded of the gingiva upto themucogingival junction in both the jaws. Grade I mobility of all the remaining teeth was present without any segmental mobility.

FIGURE 2 - A& B – Devitalized Bone and Unhealed Sockets In Both Upper And Lower Jaw.

Based on the history and clinical findings, a provisional diagnosis of ORN was given. And the other possibilities like radiation induced secondary tumour, chronic osteomyelitis and Bisphosphonate related Osteonecrosis of the Jaw (BRONJ) were considered as the differential diagnosis.

Occlusal radiographs showed unhealed sockets of 11, 12, 21, 22, 31 & 32, with mildly altered trabecular pattern in the surrounding periosteum of missing teeth. (Figure 3)

FIGURE 3 – Occlusal Radiographs

OPG also revealed unhealed sockets without any gross alteration in the trabecular pattern. There was generalized widening of periodontal ligament space without any lytic changes. (Figure 4)

FIGURE 4 – Panoramic Radiograph

Computed tomography (CT) being considered the gold standard in such cases, was also recommended. It revealed multiple areas of rarefaction in maxilla involving alveolar process, hard palate, posterolateral walls of maxillary sinus bilaterally and symphysis region of mandible. (Figure 5)

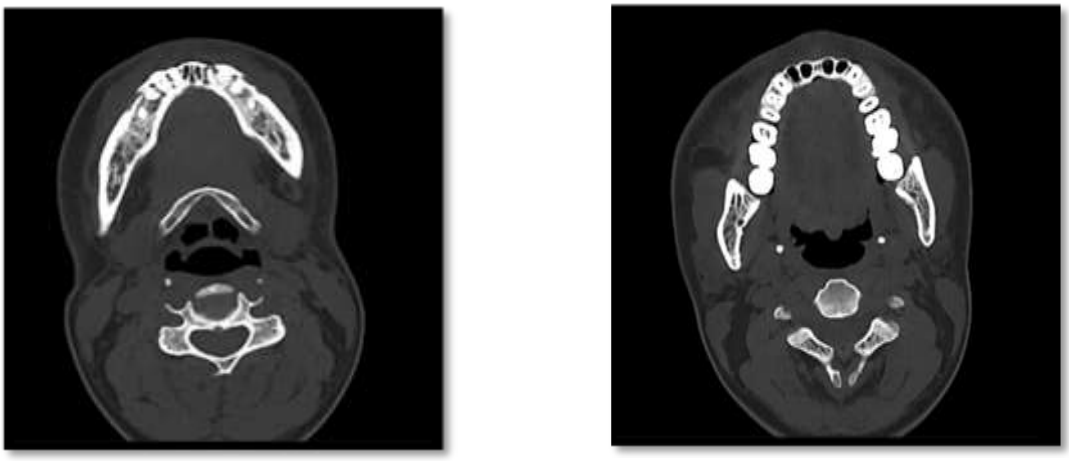

FIGURE 5 A & B – An Axial CT Image showing region of cortical bone resorption in buccal cortical plate of mandible (figure 5a), an early change in the rapeutic radiation exposure but absent in maxilla (figure 5b).

Histological analysis of the exposed bone specimen demonstrated evidence of necrotic bone fragments, accompanied by the formation of extensive fibrous tissue characterized by proliferating fibroblasts, lymphocytes, plasma cells, and an enhanced vascularity. Additionally, the specimen had a prominent presence of chronic inflammation. (Figure 6)

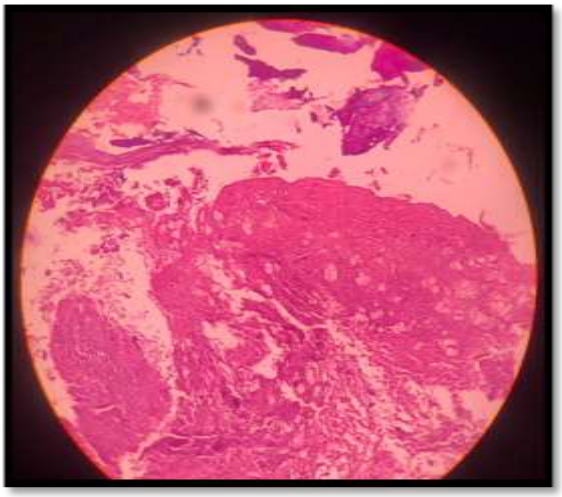

FIGURE 6 – A biopsy revealing infracted bone segments with dense chronic inflammation.

Based on the history, clinical, radiological and histopathological findings, a final diagnosis of ORN of maxilla and mandible was given.

Given the patient's history of radiation therapy, it was relatively simple to differentiate this lesion from other sclerotic lesions like chronic osteomyelitis. Furthermore, by conducting a thorough medical history review, we could rule out BRONJ as it is a well-documented complication associated with certain medications.

Patient was then referred to Department of Oral and Maxillofacial Surgery for further management.

DISCUSSION

ORN was first described by Regaude in 1922 and later by Ewing in 1926, who referred to it as radiation osteitis to encompass all the bone changes observed after radiotherapy. However, in 1983, Marx proposed the widely accepted pathogenesis paradigm known as the "3H's"for ORN. According to Marx, ORN occurs due to tissue conditions characterized by hypoxia, hypovascularity, and hypocellularity. This combination ultimately leads to tissue breakdown and non-healing wounds. Importantly, there is no infection within deep tissues; only superficial contamination exists.[6]

In 2004, Delanian and Lefaix put forth the theory that ORN is caused by a fibro-atrophic process triggered by radiation exposure. According to this hypothesis, the mechanism involves the generation of free radicals, dysfunction of endothelial cells, inflammation, micro vascular clot formation, fibrosis and restructuring of

tissues leading to eventual necrosis of bone and surroundingtissues.[7]

The head and neck region are anatomically predisposed to ORN, with the mandible being the most frequently affected bone. This can be attributed to several factors, including limited vascularity in this area, thin mucosal soft tissue coverage, mechanical stress and remodelling caused by mastication forces, as well as concurrent dental or periodontal disease. Interestingly, while the maxilla has greater porosity which promotes better blood supply, it is still commonly impacted by ORN as the second most affected region within the head and neck.[8]

The prevalence of ORN in the head and neck area ranges from 2% to 22% among individuals who have undergone radiotherapy treatment.[9]The incidence is significantly higher, about three times more, in dentate patients compared to edentulous patients. This increased risk primarily stems from complications caused by dental extractions and infections related to periodontal disease.[2]

Numerous factors have been identified as potential contributors to the occurrence of ORN. These include the dosage of radiation received (typically exceeding 60 Gy), volume of irradiated bone, type of radiation therapy used, administration of chemotherapy alongside radiotherapy, extent and size of the tumour being treated, dental extraction procedures conducted prior to or after irradiation, previous infections in oral tissues, inadequate oral hygiene practices, malnutrition status, and alcohol or tobacco abuse. Additionally, co-existing medical conditions such as hypertension, diabetes and connective tissue disorders seem to heighten the risk for developing ORN; however, the precise mechanisms through which these co-morbidities affect disease development remain unclear.[8]

In the early stages, individuals may be asymptomatic. The primary diagnostic characteristic is the exposure of bone in the mucosa or skin, which can become infected. Additional signs to look out for include pain, abnormal sensations (dysesthesia), bad breath (halitosis), and altered taste perception (dysgeusia). In more severe cases, fistulas may develop from either the oral mucosa or skin, and pathological fractures can occur.[10]

In contrast to the prevailing scientific literature, this specific case report observed greater extent of necrosis in the maxilla as opposed to the mandible. Moreover, within a period of 6 months, spontaneous exfoliation was witnessed in the affected area.

The diagnosis of ORN is established by considering the patient's medical history and identifying exposed bone that has persisted for over three months. To evaluate the extent of bone alterations, diagnostic procedures like panoramic radiographs and computed tomography are commonly employed.

There have been several suggestions from different authors regarding classification systems which consider various parameters and treatment approaches. The initial classification proposed by Marx is widely recognized, although it does have some limitations since it relies on the clinical response to hyperbaric oxygen therapy. Subsequently, Schwartz and Kagan introduced a classification that is based on clinical evaluation and physical observations rather than solely relying on HBO therapy as a determining factor.[11]

The standard treatment for ORN involves a combination of surgical procedures, oral hygiene maintenance with antiseptic products, and the administration of various medications including analgesics, antibiotics, and corticosteroids. HBO therapy may also be employed to aid in the management of ORN. In cases where conservative treatment methods have failed or when there is significant progressive disease present, more extensive interventions such as surgical resection and reconstruction may be necessary.

CONCLUSION

It is imperative to emphasize the significance of maintaining proper oral hygiene and undergoing regular dental check- ups for individuals who have undergone radiation therapy or those affected by ORN. The management of this condition requires a comprehensive approach involving the patient, general dentists, maxillofacial surgeons, as well as oncology and radiotherapy specialists. Collaborative efforts from these various disciplines are essential in providing optimal care for patients with ORN. Effective communication and coordination among healthcare professionals involved will facilitate timely intervention to address potential complications associated with this disease.

Declaration by Authors

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