Pathological fracture of the mandible associated to osteoradionecrosis: a case report

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Abstract

Objective: Osteoradionecrosis (ORN) is a severe and devastating late complication of radiotherapy in patients with head and neck cancer, especially in the mandible.

Methods: We report a case of a 47-year old woman who is suffering from osteoradionecrosis and bone exposure 5 years after irradiation therapy for squamous cell carcinoma (SCC) of the tongue. Segmented resection of the mandible was performed followed with reconstruction using a plate.

Results: ORN can cause a pathological fracture of the mandible and should be treat with a surgical approach.

Conclusion: ORN affects the mandible more often than the maxilla or any other bones of head and neck because it has high density and low blood vasculature that can cause pathological fracture of the mandible. Preventive measures must be evaluated to reduce the risk or severity of ORN.

Keyword: Osteoradionecrosis, Mandible, Pathological fracture, Radiotherapy


Introduction

Radiotherapy (RT) is usually used for head and neck malignancies as primary therapy or adjuvant to surgery, in conjunction with concurrent chemotherapy or as palliative treatment for late stage and unresectable tumours. However, high doses of RT in large areas, including the oral cavity, maxilla, mandible, and salivary glands, may result in several undesired reactions, of which osteoradionecrosis (ORN) is probably the worst.1

Osteoradionecrosis (ORN) occurs when the irradiated bone becomes devitalised and exposed through the overlying skin or mucosa, persisting without healing for 3 months in the absence of tumour recurrence.2 There is a general consensus, however, about the clinical presentations of ORN, which are pain, drainage, and fistulation of the mucosa or skin that is related to exposed bone in an area that has been irradiated. Once ORN is recognised, it is irreversible and extremely difficult to treat.3 ORN has also been described as radiation osteitis, radio-osteonecrosis, radiation osteomyelitis, osteomyelitis of irradiated bone, osteonecrosis, radio-osteomyelitis, septic osteoradionecrosis, and post-radiotherapy osteonecrosis.4

The incidence of ORN after radiotherapy for head and neck cancers has been reported to be due to the loss of soft tissue, which naturally recovers, and the exposure of necrotic bone for over 6 months. The prevalence rate also varies widely, from less than 1% to as high as 30%, with a range of 10% to 15% reported in most literature. ORN affects the mandible more often than the maxilla, with an incidence between 2% and 22%.1,4

A number of studies have suggested that the posterior portion of the mandibular body is the most common area affected by ORN because of its compact makeup and poor vascularity when compared with the anterior mandible.1,2,5 While the fractures occurred in the posterior mandible, 75% presented with pathologic fractures at the angle of the mandible. This is not surprising given the high number of mandibular fractures, including those of pathologic and traumatic origin occurring at the mandibular angle. Unfortunately, fractures located in this area are also associated with the highest rates of complications. Finally, a significant portion of pathologic fractures present without previous signs or symptoms of ORN, which is in stark contrast to its classic presentation of necrotic bone exposed through the mucosa.2,6

Several factors that are associated with risk of bone injury or ORN include patient factors (deep periodontitis, bad oral hygiene, alcohol and tobacco abuse, bone inflammation, immune deficiencies, malnutrition, and dental injury or extraction after RT), tumour factors (tumour size, stage, proximity of tumour or nodes to bone, tumour location), and treatment factors (pre-irradiation bone surgery, surgical handling of bone or its vascular supply, RT dose, biologically effective dose, photon energy, brachytherapy dose rate, combination of external beam irradiation and interstitial brachytherapy,
CASE REPORT

Case Report

Figure 1A-1C we report a 47 year old female patient who presented to the Oral and Maxillofacial Department outpatient clinic at Hasan Sadikin General Hospital. She complained about mobility of her teeth, as well as pain and swelling of her left jaw region. Five years previously, the patient had a lesion on the left side of her tongue and was diagnosed with squamous cell carcinoma by the Oncology Department at Hasan Sadikin General Hospital. She underwent combination therapy with thirty-three rounds of radiotherapy and eight rounds of chemotherapy. There was no history of trauma. From clinical examination, hyperemia of intraoral gingival, mobility of the teeth, drainage of orocutaneous fistula at left submandible region, and instability of her left jaw were observed. Figure 2 the panoramic x-ray, a discontinuity of the bone was seen at the left body of the jaw. Therefore, the patient with a pathological fracture of the jaw that was associated with osteoradionecrosis.

Following diagnosis, figure 3A-3D a multiple teeth extraction and segmental resection of the jaw was performed followed by reconstruction with an AO titanium plate.

Figure 4A-4D after the operation the wound didn’t healed well and there was a dehiscence and pus. After 10 days postoperation, it became a fistula intraoral trough the extraoral at left submandible region with diameter of about 2 cm in size.

The patient received antibiotics and open wound treatment, and the fistula became shrunk to about 0.5 mm in size 20 days after operation but there was exposure of the titanium plate 30 days postoperation figure 5A-5B.

At this point, the titanium plate was removed and a reconstruction of the fistula performed under general anaesthesia figure 6A-6B.

The wound of the incision was healing well, and the suturing was removed after 7 days postoperation. There was a little scar in the line of incision and the patient complained that she couldn’t open her mouth as wide as before. A consultation with the patient, psychologist, and rehabilitation department was arranged for mouth opening training and then the patient was scheduled for free fibular flap reconstruction figure 7A-7C.

Discussion

Pathological mandibular fractures are rare, accounting for fewer than 2% of all fractures of the mandible. They usually follow surgical interventions such as third molar removal or implant placement, result from regions of osteomyelitis, osteoradionecrosis, and bisphosphonate-related osteonecrosis of the jaw, idiopathic reasons or be facilitated by cystic lesions, benign, malignant, or metastatic tumours.

Patients with pathologic mandibular fractures related to ORN are classified as advanced ORN. Pathologic fractures in patients with ORN requires advanced therapies, with resection of the necrotic bone. The most appropriate treatment for this pathology constitutes radical intervention, removing the necrotic bone until healthy bone, with or without reconstruction.

The options for reconstruction include the use of reconstruction plate alone, reconstruction plate with free primary bone graft, reconstruction plate with secondary bone graft, or reconstruction with microvascular graft associated with hyperbaric
The microvascular reconstruction allows extensive excision of all necrotic and scarred tissue, and improves the chances of healing and achievement of healthy tissue. It also introduces tissue with a blood supply that has not been irradiated.10,11

The mandible is the longest bone in head and neck region, and more common bone to be affected by head and neck irradiation due to its unique location bearing the lower set of teeth, high density, and poor vascular supply compared with other bones in this region. Vascular supply is through inferior alveolar and facial arteries. The complication risk is highest in the region of premolar, molar, and retromolar trigone due to high density and low vascularity. Most cases of osteonecrosis post-radiotherapy (RT) occur within first 2–3 years after treatment, but patients remain at indefinite risk due to ongoing changes in the bone due to age, altered oral microflora, and dental infections.7

The severity of oral complications of radiotherapy ranges from superficial, slowly progressive bone erosion to pathological fracture. Patients often present with signs and symptoms of pain, drainage, fever, and fistula formation. These complications rarely occur in patients who have been exposed to radiation doses less than 60 Gy but are more common when brachytherapy is used and may be higher for concurrent chemotherapy and radiotherapy.4

There are many different staging systems for ORN that have been published for clinical treatment and research. These classifications were based on various criteria, such as soft tissue dehiscence, necrotic bone, oro-cutaneous fistula and pathologic fracture. Marx’s staging system is perhaps the most widely used and is predicated on staging ORN based on response to treatment.4

In early 1980s, Marx proposed the hypoxic-hypocellular-hypovascular theory as a new way of understanding the pathophysiology of ORN. Marx from his studies concluded that: “ORN is not a primary infection of irradiated bone, but a complex metabolic and homeostatic deficiency of tissue that is created by radiation-induced cellular injury; micro-organisms play only a contaminating role in ORN. The pathophysiological sequence suggested by Marx is: irradiation; formation of hypoxic hypocellular, hypovascular tissue and breakdown of tissue.”8

Preventive measures must be evaluated with the goal to reduce the risk or severity of ORN. Dental disease and dentoalveolar surgery, in particular dental extractions after radiotherapy, are well-established predisposing factors to ORN; the documented incidence of ORN after extractions is about 5%. Its incidence is three times less frequent.

Figure 4 A. Extraoral fistula 10 days postoperative, B. Intraoral fistula 10 days postoperative, C. Extraoral fistula 20 days postoperative, D. Intraoral fistula 20 days postoperative

Figure 5 Exposure of titanium plate, A. Anterior view, B. Inferior view

Figure 6 Removal of titanium plate, A. Titanium plate, B. The wound after operation

Figure 7 Profile patient 2 months postoperation, A. Anterior profile, B. Open mouth, C. Scar of the wound
in edentulous patients than in patients who retain their teeth, possibly as a result of the trauma associated with the need for extractions after irradiation and infection from periodontal disease. The risk of developing ORN after extractions is higher in posterior mandibular teeth with roots that lie below the mylohyoid line, and when an atraumatic extraction was not possible. ORN has also been reported to occur spontaneously. There are a number of risk factors that contribute to and are associated with the development of ORN.

However, as caries and periodontal disease are common, controversy has existed regarding whether such teeth should always be removed. Patients elected for RT need to receive intensive preventive dental treatment and it is now generally accepted that not all teeth in the high-dose irradiation field need to be extracted. The only teeth that really need to be extracted before RT are those within the high-dose field that are not restorable or have advanced periodontal involvement. Tooth extractions must be done before radiation therapy: most authors claiming that the prophylactic removal of periodontally involved dentition exposed to high doses of radiation minimizes the ORN risk. Furthermore, Beumer et al. reported that ORN associated with post-irradiation extractions more often requires radical resection than does ORN following pre-irradiation extractions (45.4% versus 11.7%).

Based on the current understanding of ORN pathophysiology, new protocols have been suggested for its prevention. All patients having dental extractions could be given eight weeks of pentoxifylline 400 mg twice daily with tocopherol 1000 IU, starting a week before the procedure. If ORN developed then they could be continued for a further 6 months with clodronate prescribed after 3 months if there has been no appreciable response. Patients with established ORN followed this regimen for 6 months; those who do not respond after 3 months were given clodronate. Patients who would be excluded are those with pathological fractures, or in whom pathological fracture seem likely such as when free vascularized composite tissue transfer has a role in severe, extensive, and long-established ORN particularly with a pathological fracture, but in patients who are deemed unfit for extensive surgery the pentoxifylline-tocopherol-clodronate regimen with the use of rigid fixation may be a viable treatment.

Conclusion
The mandible is the longest bone in head and neck region, and the most common bone affected by head and neck irradiation due to its unique location bearing the lower set of teeth, high density, and poor vascular supply. ORN can lead to intolerable pain, fracture, sequestration of devitalized bone, and fistulas. Pathologic fracture in conjunction with ORN has a relatively high incidence and treatment complication rate. The treatment of ORN is difficult and complex so preventive measures must be evaluated with a view to reducing the risk or severity of ORN.

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Conflict of Interest
The author reports no conflict of interest.

References


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