

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 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 plate.

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

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

Keyword: Mandible, Osteoradionecrosis, Pathological fracture, Radiotherapy

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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 tumors. 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) is irradiated bone becomes devitalised and exposed through the overlying skin or mucosa, persisting without healing for 3 months in the absence of tumor 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.^{1,2,18} 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.^{5,6}

Several factors that are associated with risk of bone injury or ORN include patient factors (deep parodontitis, bad oral hygiene, alcohol and tobacco abuse, bone inflammation, immune deficiencies, malnutrition, dental injury or extraction after RT), tumor factors (tumor size, stage, proximity of tumor or nodes to bone, tumor 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,

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field size, fraction size, volume of the mandible irradiated with a high dose).⁷

Case Report

We report a 47 years old female patient who come to Oral and maxillofacial Department outpatient clinic at Hasan Sadikin General Hospital. She complain about mobility of her teeth, pain and swelling of her left jaw region. Five years ago the patient has lesion on her left tounge and was diagnosed with Squamous Cell Carcinoma by the Oncology Department at Hasan Sadikin General Hospital. She undergone combination therapy with thirty three times of Radioteraphy and eight times



Figure 1 Profil of the patient, A. Anterior view of the patient, B. Fistula orocutaneous, C. Intraoral view



Figure 2 Panoramic x-ray



Figure 3 Durante operation, A. Segmental resection of the jaw, B. Titanium plate reconstruction, C. Suturing intra oral, D. Drainage of the wound



Figure 4 A. Ekstraoral fistula 10 days post operative, B. Intraoral fistula 10 days post operative, C. Ekstraoral fistula 20 days post operative, D. Intraoral fistula 20 days post operative



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 Profil patient 2 month post operation, A. Anterior profil, B. Open mouth, C. Scar of the wound

of Chemoteraphy. There was no history of trauma. From our examination, clinically we found hyperemism of intra oral gingival, mobility of the teeth, there was drainage of orocutaneous fistula at left submandible region and unstability of her left jaw. From the panoramic x-ray, we found discontinuity of the bone at the left body of the jaw, so we diagnose the patient with pathological fracture of the jaw that associated with osteoradionecrosis.

Then we did multiple teeth extraction and segmental resection of the jaw followed with reconstruction with AO titanium plate.

After the operation we found that the wound didn't healed well, there was a dehiscence and pus. After 10 days post operation it became a fistula intra oral trough the ekstra oral at left submandible region with diameter about 2 cm in size.

The patient recieved antibiotic and open wound treatment, and the fistula becoming smaller about 0.5 mm in size 20 days after operation but then we found exposure of the titanium plate after 30 days post operation.

So we decided to remove the titanium plate and did the reconstruction of the fistula under general anaesthesia.

The wound of the incision was healing well, and the suturing was removed after seven days post operation. There was a little scar in the line of incision and the patient complain that she couldn't open her mouth as wide as before, so we consult the patient to physic medical and rehabilitation department for mouth opening training then was scheduled for free fibular flap reconstruction.

Discussion

Pathological mandibular fractures are rare, accounting for fewer than 2% of all fractures of the mandible. They usually may 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 tumors. Patients with pathologic mandibular fractures related to ORN are classified as advanced ORN. Pathologic fracture 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.⁹

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 oxygenation therapy prior to surgery and afterwards.⁹ 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 commonest bone 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. 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 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 a view to reducing 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 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.^{lyon} There are a number of risk factors that contribute to and are associated with the development of ORN.^{3,4,8}

However, as caries and periodontal disease are common, controversy has existed regarding whether such teeth should always be removed. Patients elected to 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 unrestorable 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-radiation extractions (45.4% versus 11.7%).⁹

Based on the current understanding on 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 follow this regimen for 6 months; those who do not respond after 3 months are given clodronate. Patients who would be excluded are those with pathological fractures, or in whom pathological fracture seem likely such as when free vascularised composite tissue transfer is planned in the short term. Patients who would have been given HBO before and after curettage or sequestrectomy should be given pentoxifylline and tocopherol.³

The role of infection in ORN is not clear, particularly as the transition between ORN and osteomyelitis is ambiguous, but recent reports have suggested that bacteria may have an important role in its pathogenesis. Although the success of the pentoxifylline with tocopherol regimen seems to be successful, it is appropriate to give a short course of oral antibiotic for any extractions. Antibiotics should be used only for established ORN when there is clinical evidence of infection and frank pus, including discharging sinuses or collections. Free 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.^{1,3,4,8}

Conclusion

The mandible is the longest bone in head and neck region, and commonest 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.

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