CASE REPORT

Pathological Mandibular Fracture Secondary to Osteoradionecrosis: A Case Report

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ABSTRACT

Treatment of malignancy in the maxillofacial area includes surgery, chemotherapy, radiation therapy, or a combination of these modalities. A key component of the treatment of cancer is radiation therapy. The high doses of radi-ation used in radiation therapy drastically reduce the vascularity and repair ability of bone tissue. The most dreaded complication of radiation therapy in the jaw is osteoradionecrosis. Osteoradionecrosis of the jaws clinically shows exposed areas of bone. In advanced stages, pathological fractures of the jaws may occur. The case describes a man-dibular pathological fracture in a male patient with a history of nasopharyngeal cancer who had undergone radiation therapy 33 times with a history of tooth extraction in his upper and lower jaw. The patient was treated with segmental resection and plate reconstruction by a maxillofacial surgeon at RSUP Dr. Hasan Sadikin Bandung.
Malaysian Journal of Medicine and Health Sciences (2024) 20(SUPP12 219-223. doi:10.47836/mjmhs.20.s12.34

Keywords: Osteoradionecrosis, radiation therapy, pathological fracture of mandible,nasopharyngeal cancer, segmental resection

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INTRODUCTION

Globally, head and neck cancers (HNCs) rank seventh in terms of frequency of cancer. The pharynx, larynx, sinuses, and related areas are all involved in HNC. Comprehensive sequence therapy, which combines surgery, chemotherapy, and radiation, has proven to be a beneficial treatment for HNC. Whether used in an adjuvant or definitive setting, radiotherapy, commonly known as radiation therapy (RT), is a fundamental treatment option for HNC. Radiation therapy is used to treat about 75% of patients. Generally, there are two types of radiation therapy, internal and external (also termed brachytherapy). Using high energy photons produced by a linear accelerator, external beam radiation treatment is used to treat majority of head and neck tumors. Mechanistically, radiation damages DNA in tumor cells, but it also damages normal cells in the radiation field. Radiation can damage the DNA of the normal cells around it, but it can also alter the surrounding tissue, which over time can result in hypovascularity and less blood and oxygen reaching the affected area. There are several treatment-related late adverse consequences that can happen while treating HNC with radiation. Mucositis, changed taste, and xerostomia are examples of short-term consequences. Radiation fibrosis of the masticatory muscles, which causes trismus, is one of the longer-term consequences. Radiation caries is also commonly seen with longstanding xerostomia. The most feared side effect of radiation therapy to the jaws at high enough doses is the development of osteoradionecrosis (ORN). A non-healing, progressive necrotic process of the bone that develops over a period of more than three months following high-dose radiation therapy is indicative of a clinical diagnosis of ORN, with no probability of tumor return (1,2).

Osteoradionecrosis is the term for exposure to devitalized, irradiated bone that, in the absence of a localized malignant condition, does not heal after three to six months. Radiographically, it is characterized by thickening of the soft tissue, cortical discontinuity, lytic regions, decreased bone density, and occasionally pathological fractures. Since the mandible has a different blood supply and anatomical structure than

other regions of the facial skeleton, it is more frequently impacted by ORN. Mandibular ORN is believed to occur in two different patterns and is significantly more prevalent than maxillary ORN. About 60% are regarded as posttraumatic, whereas the remaining 40% are assumed to be spontaneous in character. Radiation results in tissue hypoxia, hypocellularity, and hypovascularity as well as endarteritis. Moreover, it decreases the growth of collagen, periosteal, endothelial, and bone marrow cells. The three-phase mechanism known as the radiation-induced fibrosis theory was developed to explain the molecular processes that result in the disease's clinical signs. Endothelial cells and an acute inflammatory response are present during the early pre-fibrotic phase. The next stage is known as the constitutive organized phase, which is characterized by a loss of extracellular matrix organization and aberrant fibroblastic activity. The last stage of the fibro atrophic phase is characterized by tissue remodelling and the development of fragile, healing tissues that are prone to recurrence. Histologically, the edges of the bones in ORN show a clear loss of osteocytes and a lack of osteoblasts. Patients may experience exposed bone in their mouths, which could lead to the loss of teeth and other supporting tissues (2,3).

Patients with ORN often arrive with complaints of poorly managed, ongoing orofacial discomfort and persistent exposure of bone. They may also develop pathological fractures, non-union with ongoing infection, orocutaneous or sinonasal fistulas. In the end, these illnesses might impair their ability to speak, breathe, and swallow. One of the most severe side effects of head and neck radiation therapy is ORN of the facial skeleton. Historically, radiation exposure, trauma, and infection were believed to be connected to it. Osteoradionecrosis of the mandible can progress to the inferior border and result in a pathologic fracture. Pain, infection, and one or more of the following conditions may be present in patients with exposed and necrotic bone, or a fistula that probes to bone: pathologic fracture, extra-oral fistula, oral antral/oral nasal communication, osteolysis extending to the inferior border of the mandible of sinus floor, or exposed and necrotic bone extending beyond the region of aveolar bone (i.e., inferior border and ramus in the mandible, maxillary sinus and zygoma in the maxilla) (1,2).

This case report aims to describe the clinical findings, modalities, and treatment of mandibular fracture due to osteoradionecrosis in patient with head and neck cancer who have undergone RT. This case report will also explain the pathophysiology and causes of ORN as well as complications of RT on oral cavity so that the clinicians will have a better understanding of this case.

CASE REPORT

A 67 years old male patient came with complaint of pain on his lower jaw and eating difficulty. +/- 7 years prior to admission, the patient was diagnosed with nasopharyngeal cancer and he had undergone a radiotherapy. +/- 2 years prior to admission, the patient complaint of pain at his right lower jaw region, then he went to a private hospital at Cimahi area but nothing was performed there. +/- 2 months prior to admission, the patient complaint of bleeding from his mouth, then he went to Emergency Department at a private hospital at Cibabat area and was performed panoramic x-ray, revealed a fracture at his right lower jaw region. Then the patient was referred to Hasan Sadikin General Hospital Oral and Maxillofacial Surgery (OMFS) Department for further treatment. +/- 2 weeks prior to admission, the patient went to Hasan Sadikin General Hospital OMFS Dept, and was performed head CT-Scan. From history taking, there was history of upper and lower tooth extraction about 3 years post RT. There was history of controlled hypertension, and coronary artery disease and was underwent a cervical reticulopathy surgery +/- 1 year ago. He's been taking Mecobalamin 500 mcg and Pregabalin 75 mg, Sodium Diclofenac 50 mg, and Amlodipine 5 mg regularly. The patient had undergone a radiation therapy for 33 times at Hasan Sadikin General Hospital about 7 years ago.

Extra oral examination of the face showed asymmetrical face and a slight oedema at right lower jaw region. On intra oral examination there was necrotic bone exposed at tooth 46 region and palpable fracture at the right body of mandible region. There were multiple radices and multiple caries found in the patient's upper and lower jaw (Fig. 1).

On panoramic x-ray, there was radiolucency on right body of mandible with irregular shape and ill-defined border from superior alveolar of mandible at tooth 46 region extended to inferior border of mandible. The extension of the lesion causing a pathological fracture (Fig. 3). On Head CT-Scan without contrast, there were impression of destruction of the right body of mandible suspected mandibular osteonecrosis (Fig. 2). The patient was diagnosed with Pathological fracture at right lower jaw region due to osteoradionecrosis, multiple caries, and Nasopharingeal Cancer stage IV.

Treatment in this case was segmental resection with reconstruction using AO plate and extraction of radices and decayed teeth on indication of pathological fracture at right lower jaw region due to osteoradionecrosis (Fig.3). Quick fixes were placed on both maxilla and mandible of the patient for intermaxillary fixation anchorage (Fig 3). The patient signs an informed consent agreement for treatment measures and approval to publish in scientific activities.

The post operation anatomical pathology concludes a result consistent with osteoradionecrosis of mandible.

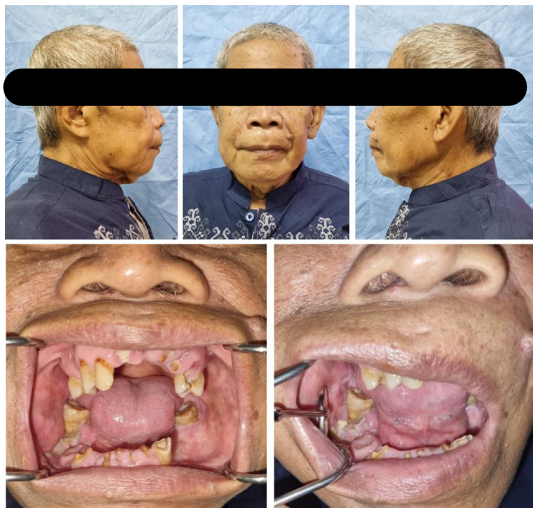

Fig. 1: Extraoral and intraoral findings.

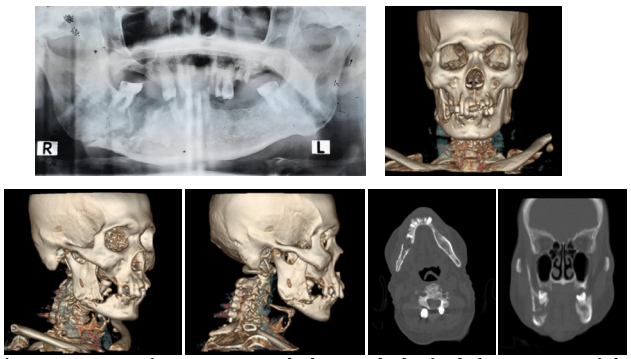

Fig. 2: Panoramic x-ray revealed a pathological fracture at right lover mandible region and head CT-Scan without contrast showed destruction on the right body of mandible

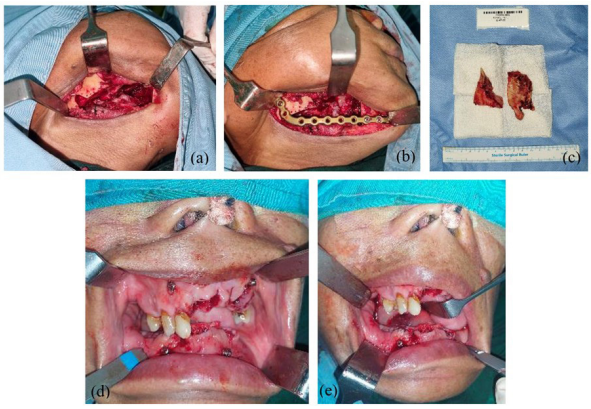

Fig. 3: Segmental mandibular resection extraoral approach with AO plate reconstruction (a,b) Specimen of resected necrotic mandible (c) Intraoperative figure shows the installation of quick fix on both maxilla and mandible bilateral (d,e)

DISCUSSION

Nasopharyngeal malignancies are often difficult to treat and necessitate a multimodal approach that includes radiation, chemotherapy, and surgery (3). In this case the patient was suffered nasopharyngeal cancer and was treated with radiation therapy and had undergone 33 times of radiation. A small percentage of individuals undergoing radiation therapy for head and neck malignancies may get jaw osteoradionecrosis. Individuals who suffer from osteoradionecrosis of the jaw may experience exposed mandibular bone intraorally. While many of these individuals may go on to heal naturally and without any complications, some may get osteomyelitis or even mandibular fractures. The patient in this case was a 67 years old male who had suffered a nasopharyngeal cancer for 7 years. According to West et al, typically, individuals with osteoradionecrosis are older than 55 (4). It is believed that radiation's harmful effects on blood vessels and suppression of neovascularity are the secondary causes of ORN development. The technique works mechanistically by directing ionizing radiation toward diseased tissue, which damages the tissue. Free radicals produced by the ionizing radiation led to genetic degradation that eventually leads to tumor cell death and a lack of cellular reproduction. While normal cells are often more resilient to the effects of ionizing radiation, even those that do may have compromised collagen and regulating enzyme production processes. In the end, this causes a gradual loss of tissue integrity, cellularity, and vascularity (1).

Necrosis of the bone results from these processes, making it more vulnerable to co-morbid infection and osteomyelitis development. Although patients with osteomyelitis may also have necrotic bone, those with radiation history may have significant fibrotic replacement of marrow in addition to a more subtle clinical course that sets ORN apart from acute infection alone. Endothelial cells and an acute inflammatory response are present during the early pre fibrotic phase. The next stage is known as the constitutive organized phase, which is characterized by a loss of extracellular matrix organization and aberrant fibroblastic activity. The last stage of the fibro-atrophic phase is characterized by tissue remodelling and the development of fragile, healing tissues that are prone to recurrence. Osteocytes have clearly been destroyed in ORN, and osteoblasts are not present in the borders of the bones. The fibro-atrophic process that follows bone irradiation and the ensuing hypovascularity and hypocellularity appear to play a critical role in the etiology of the ORN, creating delicate tissues prone to necrosis, particularly in situations when tissue injury has occurred, like in tooth extractions. In this instance, the patient had a tooth extracted roughly three years following radiation treatment. After radiotherapy, tooth extractions are acknowledged as the primary risk factor for the beginning of ORN, with reported incidences varying from 2% to 22% of patients, depending on the various populations studied and diagnostic measures used (2,5).

The patient was suffered a pathological fracture of the mandible. Davis et al (2023) has described the severity of the mandibular ORN with Robert Marx's staging system and treatment protocols. In Stage I, a rapidly developing chronic exposed bone without pathological fracture necessitated preoperative hyperbaric treatments and bony debridement, while in Stage II, not responsive to hyperbaric therapy and requires sequestrectomy

and saucerization. Stage III requires resection and reconstruction of mandibula due to pathologic fracture, percutaneous fistulae, and lytic lesion that extend to the inferior border of the mandible. According to Vahidi et al (2020), surgery has continued to be a mainstay of treatment for the mandibular ORN. Surgery is usually recommended for patients who have not responded to the proven medical treatments such as medical management and hyperbaric oxygen treatment, or who have shown signs of serious ORN complications. Among these issues are mandibular pathological fractures and significant soft tissue defects that reveal metal or bone. The precise surgical course of action will vary depending on the number of variables and needs to be customized for every patient. Among these are the extent of exposed mandibular bone, the location of the bone exposure along the mandible, the depth of the bony defect (partial vs. full-thickness mandibular defect), the quality of the surrounding soft tissue that will be used for reconstruction, and the existence of intraoral mucosal defects or concurrent facial skin defects that will also require reconstruction.

As of right now, ORN lacks a generalized consensus treatment algorithm. Surgical treatment aims to replace any exposed hardware and cover it with vascularized tissue to prevent biofilm formation and infection, which can lead to resistance to antibiotics, including intravenous therapy and provide stiff fixation in the event of a post radiation pathological fracture to facilitate bone union. When there is a segmental bone defect, use stiff fixation to restore mandibular continuity and premorbid occlusion. Use vascularized tissue to reconstruct contemporaneous intraoral mucosal and/or skin defects. When there are significant bone defects from a segmental mandibulectomy combined with contemporaneous intraoral and/or external skin abnormalities, restoration may require the use of a vascularized osteocutaneous free flap. Remove any compromised teeth when having the final treatment procedure done. To prevent future infection, it is advisable to extract the affected tooth and aggressively debride the alveolar bone until a bleeding bone edge is detected if there are indications of dental infection within 1 cm of the ORN site (2,3).

Treatment in this case was segmental resection with reconstruction using AO plate and extraction of radices and decayed teeth on indication of pathological fracture at right lower jaw region due to osteoradionecrosis. Surgical removal of all necrotic bone is required. The most utilized alloplastic devices for mandibular reconstruction are screws and plates. To provide a mandibular reconstructive option that was quick, simple, and dependable while preserving oral function and shape, AO stainless steel and AO titanium reconstruction plates were created. The effectiveness of using these plates has varied. The jaw should be lowered to its natural position before plating; any teeth that may be present should have wear facets that are suitably opposing each other to ensure that the jaw is in its pre-traumatic or pre-operative configuration. Among the techniques for achieving a temporary maxillary-mandibular fixation are arch bars, screws, and 24-gauge wires. Before reconstructing an area with comminute fractures, the wound should be irrigated and the bone fragments removed. A reciprocating saw can be used to remove an extra 1-2 mm of bone until fresh, bleeding edges are visible if a portion of the mandible is gone. This will help the healing process if the remaining bony edges are freshened. For many years, after resection, the standard therapeutic approach disregarded the condyle, leaving the ramus free to float. Despite the fact that many patients have an appropriate interincisal opening, mandibular deviation and altered functional occlusion are common. Condylar reconstruction is used to maintain preoperative occlusion, maintain a sufficient interincisal opening, and preserve the mandibular dual joint function that stabilizes the masticatory muscles (3,5).

Dental extractions performed after radiation therapy are believed to be one of the most frequent causes of ORN, with an incidence that ranges from 2 to 18%. Research has indicated a rising incidence of ORN associated with dental extractions during the initial 4 to 5 years following radiation therapy. Therefore, it is advisable to minimize the need for extractions during this post radiation phase, ideally completing any necessary extractions before beginning radiation therapy. When it comes to tooth extractions performed before to radiation therapy, timing is also rather crucial. If time is of the essence, extractions should take place at least 3–4 weeks before radiation therapy; if this is not possible, at least 10–14 days should be allowed for recovery. The intention is to give the body enough time to recover before radiation therapy compromises it (5). Osteoradionecrosis can present with a range of signs and symptoms, including discomfort, fistulas, sequestration of necrotic bone, and more severe cases involving mandibular fractures that require significant surgical operations or result in potentially fatal infections. Patients usually complain of head and neck pain when they are symptomatic, and the duration of the radiation treatment varies, taking anywhere from a few months to several decades (3).

CONCLUSION

Osteoradionecrosis of the jaw may develop after radiation therapy for nasopharyngeal cancer therapy which causes tissue hypovascularization and hypocellularity, thereby reducing tissue reparative ability when an injury occurs or a tissue damage, including in tooth extraction cases. Sustained bone destruction can lead to pathological fractures of the jaws.

ACKNOWLEDGEMENT

The author would like to thank the supervisors who have provided the opportunity to complete this case report

REFERENCES

1. Fonseca RJ. Oral and Maxillofacial Surgery-Inkling Enhanced E-Book: 3-Volume Set. Elsevier Health Sciences; 2017 Mar 8.
2. Vahidi N, Lee TS, Daggumati S, Shokri T, Wang W, Ducic Y. Osteoradionecrosis of the midface and mandible: pathogenesis and management. InSeminars in Plastic Surgery 2020 Nov (Vol. 34, No. 04, pp. 232-244). Thieme Medical Publishers, Inc. DOI: 10.1055/s-0040-1721759
3. Davis DD, Hanley ME, Cooper JS. Osteoradionecrosis. InStatPearls [Internet] 2022 Jun 21. StatPearls Publishing. PMID: 28613568
4. Lajolo C, Rupe C, Gioco G, Troiano G, Patini R, Petruzzi M, Micciche F, Giuliani M. Osteoradionecrosis of the jaws due to teeth extractions during and after radiotherapy: a systematic review. Cancers. 2021 Jan;13(22):5798. DOI: 10.3390/cancers13225798
5. Smith, J.E. 2023, March. Mandibular Plating. Available from: https://emedicine.medscape.com/article/881542-overview