

Mandibular Reconstruction with Deep Circumflex Iliac Artery Osteomyocutaneous Free Flap in a Severe Case of Osteoradionecrosis of the Mandible: A Case Report

Namkabir Singh^{1*}, Kong Chee Kwan², Siti Mazlipah Ismail¹, Alizan Abdul Khalil²

¹Department of Oral & Maxillofacial Clinical Sciences, Faculty of Dentistry, Universiti Malaya, 50603 Kuala Lumpur, Malaysia
²Department of Plastic & Reconstructive Surgery, Faculty of Medicine, Universiti Malaya, 50603 Kuala Lumpur, Malaysia

ARTICLE INFO

Article history:

Received 10 July 2023
Revised 01 October 2023
Accepted 25 March 2024
Online first
Published 1st September 2024

Keywords:

osteoradionecrosis
mandible
radiotherapy
reconstruction

DOI:

10.24191/cos.v11i2.27509

ABSTRACT

Introduction

Osteoradionecrosis (ORN) can be accompanied by several signs such as tissue loss, exposed necrotic bone, presence of fistula, pain, infection, trismus, and even pathological fracture of bones. Early-stage ORN is usually managed conservatively with medication while severe progression of the disease requires aggressive treatment such as free flap reconstruction surgery.

Case report

This case involves a 52-year-old lady who was previously diagnosed with well-differentiated squamous cell carcinoma of the left side of the tongue (T3 N2b M0) and had undergone surgery, followed by 6 cycles of chemotherapy and 30 cycles of radiotherapy. She presented to our centre complaining of pain and pus discharge over her lower jaw for the past 3 months. Upon examination, the skin around her neck and chin was erythematous, firm and tense, limiting not just her mouth opening but also the flexibility of her neck movement. She also had two oral-cutaneous fistulas, at the submental and submandibular region, with thick pus discharge. Intraorally, there was a broken and exposed titanium plate. She was subjected to segmental resection of the anterior mandible together with fistulectomy, reconstruction with deep circumflex iliac artery osteomyocutaneous flap and tracheostomy.

Conclusion

Despite not being a life-threatening disease, ORN has a significant impact on the well-being of patients, leaving them with possible disfiguration, pain and chronic wounds. This case report summarizes the journey of our patient from initial presentation, surgery, and post operative follow-up, highlighting the rare need for aggressive treatment in managing advanced ORN.

^{1*} Corresponding author. E-mail address: namkabir1987@gmail.com

INTRODUCTION

An area of exposed, non-vital bone that has previously undergone radiotherapy with inadequate healing and remodelling over a period of 3 to 6 months without the presence of local neoplastic disease is defined as osteoradionecrosis (ORN) (Vahidi et al., 2020). It can also be described as radiation-induced ischemia of the bone and soft tissues with varying extension and delayed healing without the presence of tumour recurrence or metastatic disease (De Lima Dantas & Reis, 2019). The mandible is the most frequently affected facial bone due to its anatomic structure and blood supply. Despite posing a life-long risk of acquiring ORN, it is estimated that up to 94% of ORN cases appear within the first 3 years of radiotherapy (Vahidi et al., 2020). Studies have shown that between 5% to 10% of patients undergoing radiotherapy of the jaw end up developing ORN as a complication (Tymofieiev & Tymofieiev, 2018). ORN usually occurs due to a traumatic event despite having the ability to appear spontaneously (De Lima Dantas & Reis, 2019).

Complications associated with ORN include pain, trismus, extraoral fistula formation, purulent discharge, non-healing ulcers, sequestrum formation, malocclusion, food impaction and fetor oris. Some patients even develop neurological symptoms such as dysgeusia, dysesthesia or anaesthesia. Progression of ORN often leads to pathological fracture of jawbones, local or systemic infection, disturbance in speech and difficulty in mastication. There have been multiple different staging systems that have been proposed by various researchers over the years. The current most adopted classification is by Notani et al. in 2003, in which ORN is divided into 3 stages. Stage I is when ORN is limited to the alveolar bone only, whereas stage II occurs when ORN is limited to the alveolar bone and the mandible above the level of the inferior alveolar canal. Stage III on the other hand is defined as ORN that involves the mandible below the level of the inferior alveolar canal, with or without skin fistula and pathological fracture (Chronopoulos et al., 2018). Treatment for stage I and stage II usually include close observation, meticulous oral hygiene, mouth rinses that contain chlorhexidine, antibiotics for acute infection, analgesics as needed, and removal of local irritants and ill-fitting dentures. Other interventions such as sequestrectomy, wound debridement and trimming of sharp bony edges are also employed in early stage ORN. As stage III ORN is advanced disease that leads to pathological fractures and fistula formation, surgical intervention such as resection and reconstruction using osteo cutaneous free flap is generally employed. Hyperbaric oxygen is frequently used as an adjunct to treat ORN. It increases the oxygen gradient by enhancing the diffusion of oxygen into hypoxic tissues. However, until now literature supporting the effectiveness of hyperbaric oxygen in treating ORN has remained controversial (Singh et al., 2022).

CASE REPORT

A 52-year-old lady presented to our centre complaining of pain and pus discharge over her lower jaw. Her first symptom was mild pain in the lower jaw, which began about 3 months prior to presentation. This was followed by more severe pain several weeks later. She also noticed that there was a foul-smelling yellowish discharge from the skin at her chin area for the past 2 months. This caused her to have a lot of difficulty in chewing her food, forcing her to resort to nourishing fluid only for approximately 4 to 5 weeks before seeking treatment at our centre. At the same time, she shunned herself socially as she feared that the pus discharge from her chin would repulse people, prompting her into depression. She sought treatment for her depression at a private psychiatrist and was started on oral sertraline 25mg daily. She had also lost a substantial 5 kilograms over the past several weeks. Despite being apprehensive at first, she finally decided to seek treatment at our clinic once she noticed the exposed metal plate in her oral cavity.

Further history from the patient revealed that she was previously diagnosed with well-differentiated squamous cell carcinoma of the left side of the tongue (T3 N2b M0). Left hemiglossectomy, left modified radical neck dissection, right supraomohyoid neck dissection and subsequent reconstruction with a gracilis flap were performed on her 11 months ago at our centre as well. During the surgery, mandibulotomy was done to gain access to the tongue, and it was plated back using a titanium plate. Surgery was succeeded by

6 cycles of chemotherapy and 30 cycles of targeted radiotherapy amounting to 65 Gy. She completed her radiotherapy 8 months ago and was under close monitoring for about 3 months before defaulting to subsequent follow-ups. She was otherwise not suffering from any other ailments or known drug or food allergies. She also denied any deleterious habits.

She suffered from many side effects of radiotherapy following completion of treatment. Immediately post-radiotherapy, she had severe trismus and was only able to open her mouth to the width of 1 finger breadth. Concurrently, she also experienced xerostomia. Despite being prescribed with oral 7 mouthwash to alleviate her discomfort, she claimed that there was no improvement in her symptoms.



Fig. 1. Cutaneous fistulas at level Ia & Ib (indicated by black arrows).

Upon examination, the skin around her neck and chin was erythematous, firm and tense, limiting not just her mouth opening but also the flexibility of her neck movement. There were two oral-cutaneous fistulas present, one at level IA (submental) and another at level IB (submandibular), with thick pus discharge. Intraorally, the titanium plate was broken and exposed. At the same time, what appeared like necrotic bone was also exposed at the anterior region of the mandible. The tongue flap was otherwise pink and healthy with no evidence of recurrence. A contrast-enhanced computed tomography (CECT) neck was taken, and the results showed a pathological fracture of the symphysis of the mandible with multiple sequestrums and a broken titanium plate. Hence, the patient was diagnosed with ORN of the mandible.

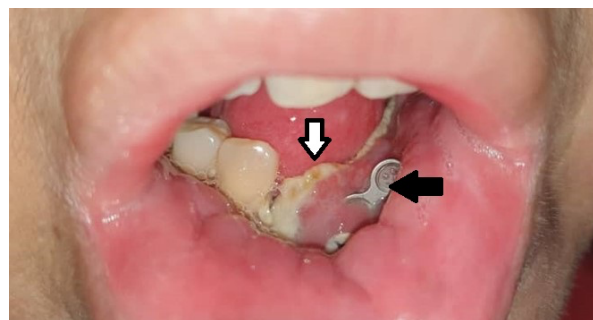


Fig. 2. Intraoral exposure of broke plate (indicated by black arrow) and fractured anterior mandible (indicated by white arrow).

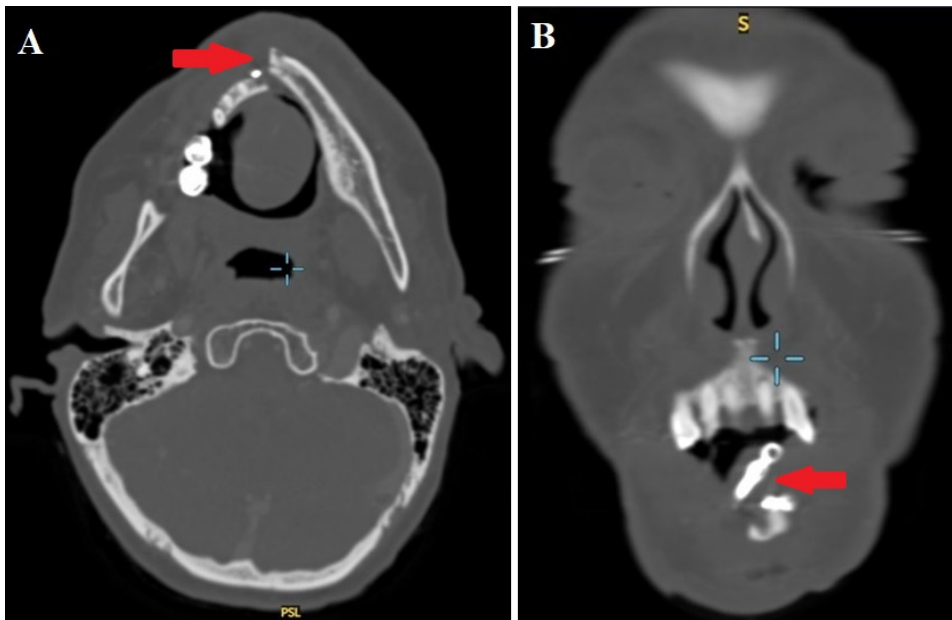


Fig. 3. CT scan findings. (A) Axial view of CT showing pathological fracture of symphysis of mandible (indicated by red arrow). (B) Coronal view showing fracture of titanium plate (indicated by red arrow).

She was then scheduled for segmental resection of the anterior mandible together with fistulectomy, reconstruction with deep circumflex iliac artery osteomyocutaneous flap and tracheostomy. At induction, 1.5 grams of Cefuroxime, 25mcg of fentanyl and 50mg of propofol were administered intravenously. During the surgery, the first step taken was to make the surgical markings. Then an incision was made along the scar from the previous incision site, extending submentally to include the two cutaneous fistulas.

Once the flap was raised, segmental resection of the anterior mandible, from the left to right parasymphysis region measuring approximately 7 cm was done to remove necrotic bone. Skin over the submental area was also resected measuring 9cm x 6cm. Subsequently an incision was made at the left hip measuring approximately 9cm x 6cm. The deep circumflex iliac artery (DCIA) was then identified and traced distally towards the anterior superior iliac spine, in the substance of the transversalis fascia. Dissection was done until the appropriate length of iliac crest bone was exposed. An oscillating saw was used to osteotomize 7cm of the iliac crest bone to replace the resected mandible. The skin paddle together with the bone was then divided and transferred to the neck region, whereby the deep circumflex iliac artery was anastomosed to the left superior thyroid artery and the vena comitants to the left internal jugular vein. Excellent inflow and outflow were confirmed. The harvested iliac bone was contoured to the dimension of the reconstruction plate and fixated towards the remaining mandible bone using the already bent 2.0mm thick reconstruction plate and screws. Then, antibiotic-loaded cement (Tobramycin) was secured to the remaining iliac bone using wires to restore the curvature of the hip. Afterwards, a tracheostomy was done. Finally, one drain was inserted at the left neck and two drains at the hip region, before wound closure was done. Post-operatively, she was admitted into the intensive care unit (ICU) for one day before being transferred out to the general ward.

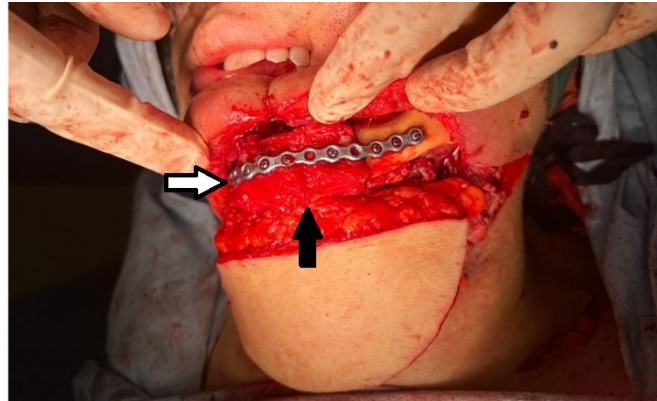


Fig. 4. Contoured iliac bone (indicated by black arrow) secured to mandible using a 2.0mm thick reconstruction plate (indicated by white arrow).

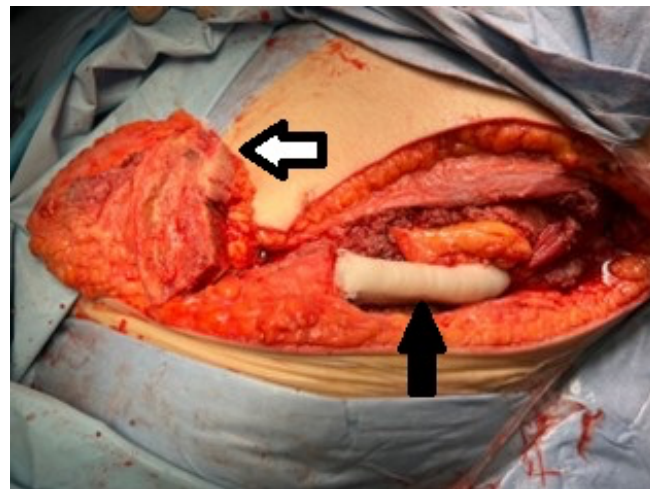


Fig. 5. Skin paddle together with iliac bone harvested (indicated by white arrow), antibiotic-loaded cement attached with wires to remaining iliac bone to recontour the hip (indicated by black arrow).

Post-operatively, she required close monitoring for the next couple of days. She was started on intravenous piperacillin-tazobactam 4.5 grams four times a day together with a combination of patient-controlled analgesia (PCA) morphine 1 mg and oral celecoxib 200 mg. She required the PCA for 3 days before being solely reliant on only oral analgesics. Her free flap was monitored hourly for the first several days to ensure adequate perfusion. The following day after surgery she was already started on nourishing fluid, which was slowly stepped up based on the high protein and high-calorie regime suggested by the dietitian. Three days after surgery, wound inspection was done over both her abdominal wound and free flap. The uptake of the free flap was good, and the flap was pink and warm with good turgor and a capillary refill time of less than 2 seconds. The abdominal wound also healed well, with no signs of wound breakdown or seroma. Her vacuum drains however were still actively draining hemoserous liquid and were maintained for a total of 6 days.

On day 4 after surgery, she had already started assisted ambulating, and only complained of pain mainly at the donor site. The recipient site continued to heal well. Seven days after surgery, the flap uptake was good and she was ambulating well around the ward. She was also tolerating clear fluid orally and complained of minimal pain at the left hip. Having already completed seven days of intravenous Tazocin

(Piperacillin & Tazobactam) 4.5 grams four times daily, she was allowed discharge home with regular follow-ups at the outpatient clinic.

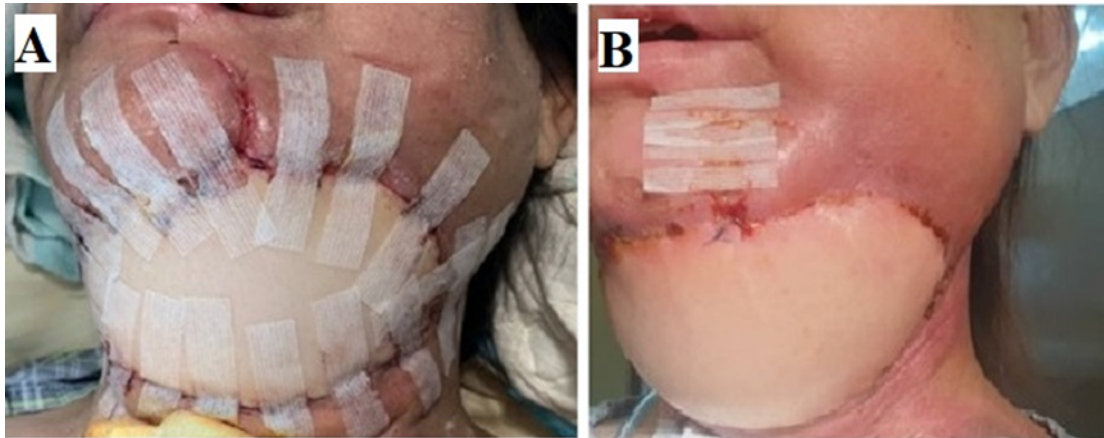


Fig. 6. Condition of the free flap. (A) 2 weeks after surgery. (B) 1 month after surgery.

During her review at the clinic one month after surgery, the patient appeared to be in good spirits as she was in much lesser pain. At the same time, her flap had healed well, with no discharge. It was pink, warm and had good turgor. Unfortunately, she did still have minimal trismus, but with a much more improved mouth opening, allowing her to tolerate a normal diet. She was then scheduled for regular follow-up at our clinic for continuation of care. However, she once again defaulted her scheduled follow-up and was unreachable when contacted.

DISCUSSION

An established complication of radiotherapy is ORN, which occurs because of the inability of radiated tissue to recover spontaneously. Radiation therapy can be divided into two, internal radiotherapy or more commonly known as brachytherapy and external beam radiotherapy which consist of intensity-modulated radiation therapy (IMRT), concurrent chemo-radiation therapy (CCRT) and conventional radiotherapy to name a few (Vahidi et al., 2020). External beam radiation is often the preferred method for head and neck malignancies. Prevalence of ORN was found to be 7.4% in conventional radiotherapy, 5.3% in brachytherapy, 5.3% in CCRT and 5.1% in IMRT (Vahidi et al., 2020). For necrosis of tissue to occur, several factors must be taken into consideration, notably the location of the radiation, the amount of skin exposed and the dose of radiation given (Tymofieiev & Tymofieiev, 2018). Studies show that the capillary density of radiated tissue is reduced to only 20% to that of healthy tissue at radiation doses of between 65 to 80 Gy. Patients who receive 40 to 60 Gy of radiation are at a medium risk of developing ORN, while those who receive more than 60 Gy are at high risk of developing ORN (Vahidi et al., 2020); (Ajila & Hegde, 2020).

ORN can be accompanied by several signs such as tissue loss, exposed necrotic bone, presence of fistula, pain, infection, trismus, and even pathological fracture of bones. Other conditions associated with ORN include xerostomia, dysgeusia, dysphagia and reduced tongue mobility. Development of one or more of these conditions frequently causes patients to be physically and emotionally distressed (Jacobson et al., 2010). Our patient experienced a similar condition, being embarrassed to attend any social event and malnourished due to her limited mouth opening, pain, and inability to chew. ORN also took a toll on her mental health by causing her to suffer from depression and requiring her to seek treatment at a psychiatric clinic for her ailment. This just goes to show the severity of the negative impact ORN can have on one's well-being.

ORN is a complex and progressive disease that is difficult to treat. For early-stage ORN, conservative management is often applied, to restrict the progression of disease. This is usually achieved via pharmacological management such as consumption of antibiotics and a combination of pentoxifylline-tocopherol or by using hyperbaric oxygen and sequestrum removal. However, severe cases require more aggressive treatment, notably free flap reconstruction surgery for replacement of necrotic and poorly vascularized tissues (De Lima Dantas & Reis, 2019). With regards to the treatment of ORN, the curative rate when applying conservative management stands at 60%. However, when all else has failed, surgical treatment represents a curative rate of approximately 40% (Spijkervet et al., 2019); (Gard et al., 2018).

Despite having undergone reconstruction of the defect with a free flap, the patient's well-being may still be affected due to the complications of radiotherapy. Hence, all treatment options must be explained and disclosed to the patient, covering not only the treatment phase but also the early and late sequelae of each treatment option. With mandibular reconstruction being a complex procedure and ORN demonstrating poor prognosis, careful planning must be carried out with importance placed on restoring the patient's function and aesthetics after treatment has been completed (Wang et al., 2009). A study was carried out by Wang et al. in 2009 on the quality of life in osteoradionecrosis patients after mandible primary reconstruction with free fibula flap. Several aspects were looked at including pain, appearance, speech, swallowing, chewing, taste, saliva, anxiety and mood. The results from the study concluded that mandible resection and reconstruction improved health-related quality of life in the patient's perception. Mandible reconstruction with a free flap also effectively eliminates pain and brings local infection under control, although radiotherapy-induced complications still adversely influence the quality of life of ORN patients even after reconstruction (Wang et al., 2009). A more recent survey conducted among ORN patients who have undergone reconstruction with free flap surgery by Sweeney et al. in 2021 reported that 45% of patients had no pain, 28% had no swallowing abnormalities and 93% had no speech difficulties. They concluded that surgery provided patients with a good return of function (Sweeney et al., 2021). Thus, even though ORN may not be a life-threatening disease, an aggressive approach is sometimes more beneficial and should be considered when dealing with severe ORN, to not only return function and restore confidence, but also to generally improve their well-being.

Challenges faced when performing free flap surgery in these patients include lack of suitable vessels in the neck, bony non-union and risk of plate infection (Urken et al., 1994). The sites that are most commonly utilized in the neck region include the facial artery, faciolingual trunk and superior thyroid arteries. However, in patients with ORN, these blood vessels may be damaged, ligated, be of insufficient length, might have been utilized in the previous free flap surgery or damaged by radiotherapy. Therefore, relying on the availability and calibre of those vessels to sustain a satisfactory blood flow remains risky (Wong et al., 2013). We were fortunate as the superior thyroid artery of our patient was suitable for anastomoses, hence the satisfactory healing of the flap. Our patient was satisfied with the outcome as she no longer suffered from pain and foul-smelling discharge. She was also able to open her mouth slightly wider and tolerate a normal diet, enabling her to continue living a normal life without any debilitation.

CONCLUSION

ORN is one of the worst complications preceding radiation for head and neck malignancies, as it leaves patients with not only chronic wounds but also limitations in function and pain. Free flap surgery remains one of the best options available for advanced ORN due to its impressive track record. Nevertheless, it does come with its fair share of complications such as post-operative wound infection and functional decline. Thus, regular communication between the practitioner and patient regarding realistic expectations and anticipated outcomes is essential in treating these complex cases.

ETHICS APPROVAL

While ethics approval is not applicable in this case report, the principles outlined in the Declaration of Helsinki were strictly followed throughout the entire process to ensure that the authors acted in the patient's best interest when providing medical care while adhering to the ethical guidelines. Written informed consent for patient information and images to be published was provided by the patient.

DECLARATION OF COMPETING INTEREST

There are no conflicts of interest to declare.

ACKNOWLEDGMENT

The authors would like to thank the patient for her willingness to share her medical records and images.

AUTHORS' CONTRIBUTIONS

NS prepared the manuscript and performed the editing. KCK, SMI and AAK performed surgery for the patient and were involved in the manuscript review and editing. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

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