



**CASE REPORT**

# Refractory ORN submitted to resective surgical treatment and free flap musculocutaneous reconstruction

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## Abstract

Osteoradionecrosis (ORN) is one of the most severe complications of radiotherapy in the head, having the greatest impact on patients' quality of life. Previous studies have shown injury remission and cure rates for refractory osteoradionecrosis, i.e. ORN resistant to conventional treatment, using simultaneous techniques such as superficial debridement, midsize sequestrectomy and resective surgery along with drug therapy and hyperbaric chamber. The present case report describes the results obtained exclusively by a resection surgical approach to ORN and microsurgical reconstruction with musculocutaneous free thigh flap.

**Keywords:** osteoradionecrosis; surgical treatment; microsurgical reconstruction; myocutaneous flap; combined therapy.

## Introduction

Osteoradionecrosis (ORN) is one of the most severe complications in patients irradiated in the head and neck region, defined as the area of exposed necrotic bone that does not heal three to six months after the first clinical detection. It is caused mainly by decreased vascularization, which triggers tissue hypoxia and loss of cellularity in the affected bone due to the direct and/or indirect effects of radiation therapy. Many surgical approaches have been employed, aiming at the complete remission of this clinical condition; however, there is still no consensus on the best treatment. Combined therapies have been used to treat refractory cases of the disease. Refractory ORN is characterized as resistant to multiple attempts at treatment, especially conservative non-surgical therapies, since these have shown a lower clinical success rate compared with surgical treatments involving broad resections<sup>1,2</sup>.

Previous studies have described combined treatments, in which non-surgical therapeutic modalities have been used jointly with surgical approaches. One of the most frequently reported approaches in the literature is hyperbaric oxygen therapy (HBOT) before and after superficial and extensive sequestrectomies, aiming at revascularization and oxygen support of tissues that remain after removal of the devitalized bone<sup>3</sup>.

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Based on the protocol proposed by Delanian et al.<sup>4</sup>, drug therapy *per se* consists in the stabilization of fibro-atrophy as a pathophysiological mechanism of the disease, using pentoxifylline as an antifibrotic and revascularizing agent; tocopherol, with high antioxidant potency; and clodronate, an anti-resorptive bisphosphonate with antiangiogenic effects and direct action on osteoblastic cells (PENTOCLO). It has shown satisfactory clinical success rates in the treatment of refractory ORN, after using the treatment for three to four months, concomitant with sequestrectomies and medium-sized debridements, as reported in the literature.

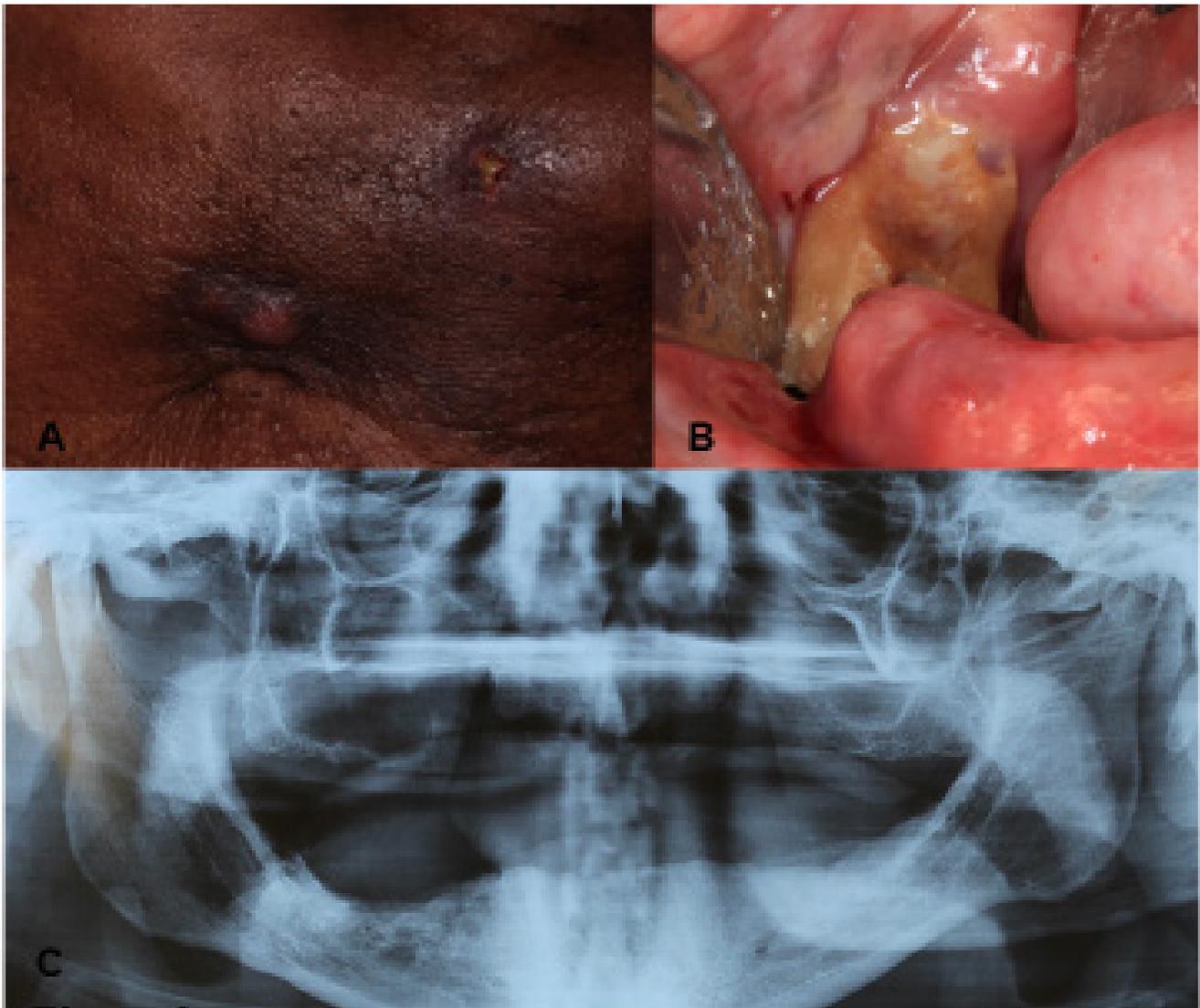
Although conservative management of ORN is adequate therapy for the treatment of earlier-stage or low-degree disease, the resection surgical approach is recommended in more advanced and refractory stages of ORN, in cases of orocutaneous fistulas, pathological fractures, extensive bone exposures, and large soft tissue defects<sup>5</sup>. Depending on the defect, reconstruction may be done using local microsurgical flaps, autogenous bone graft, and reconstruction plates and screws, as well as free tissue transfer, although free flaps vascularized with bone segments are one of the alternatives, if not the one with best results in reconstructive therapy<sup>5</sup>.

Also, surgical resection has shown favorable results in the search for complete remission of ORN, and is used in most cases or jointly with other therapies. Advanced ORN injuries, refractory or not, were treated with broad resections and reconstructions using microsurgical techniques, using grafting and free flaps taken from other donor areas such as iliac crest, fibula, and thigh<sup>5</sup>.

The present case report describes the results of a broad resection of an ORN injury whose reconstruction was carried out using the microsurgical technique with musculocutaneous thigh flap.

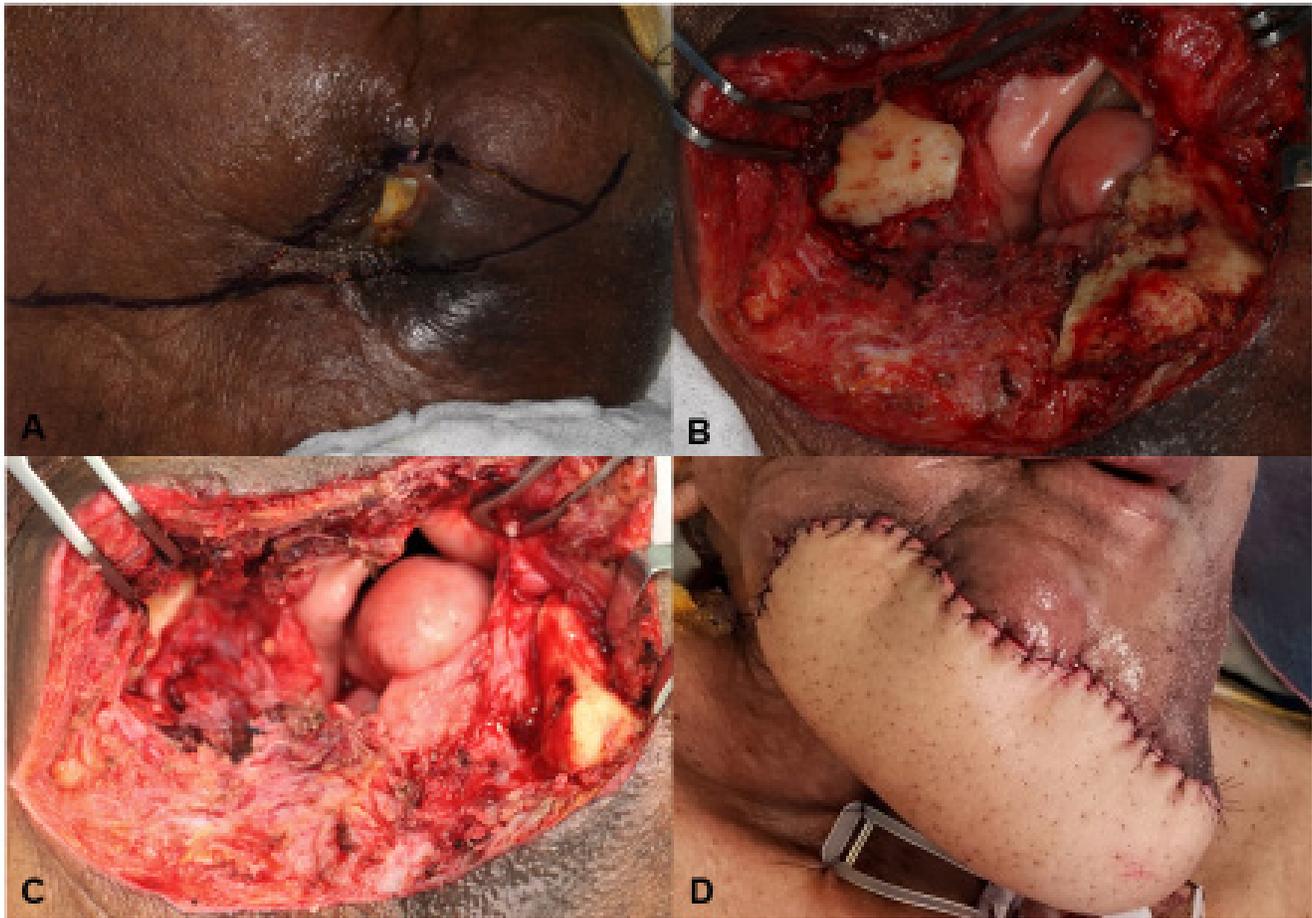
### Case report

A 64-year old male patient with a history of oropharyngeal squamous cell carcinoma (CPB) as an underlying disease underwent radical surgical treatment consisting of hemiglossectomy in combination with right-sided neck dissection and local adjuvant radiotherapy. Radiotherapy treatment consisted of 200cGy delivered fractionally in 30 sessions between April and May 2013. After 5 years of completion of treatment for the primary tumor, the patient presented to the Stomatology Dept. of A.C. Camargo Cancer Center in São Paulo, Brazil, for evaluation of the orocutaneous fistula in the right hemi-face region. Bone exposure was observed (approximately 2.5cm long, in the anterior region of the right mandibular body) during clinical examination, and radiographic assessment revealed osteolytic areas compatible with bone sequestration regions, as shown in Figure 1. With a clinical and radiographic diagnosis of mandibular ORN, the patient underwent conservative surgical debridement of the suppurative osteolytic lesion using the intraoral approach via a mucoperiosteal incision with removal of the devitalized bone tissue. Subsequently, a thorough curettage and surgical field washing were done with primary closure of the covering mucosa. The initial diagnosis of osteoradionecrosis was confirmed after the anatomopathological analysis of the surgical specimen. Six months later, the patient went to the



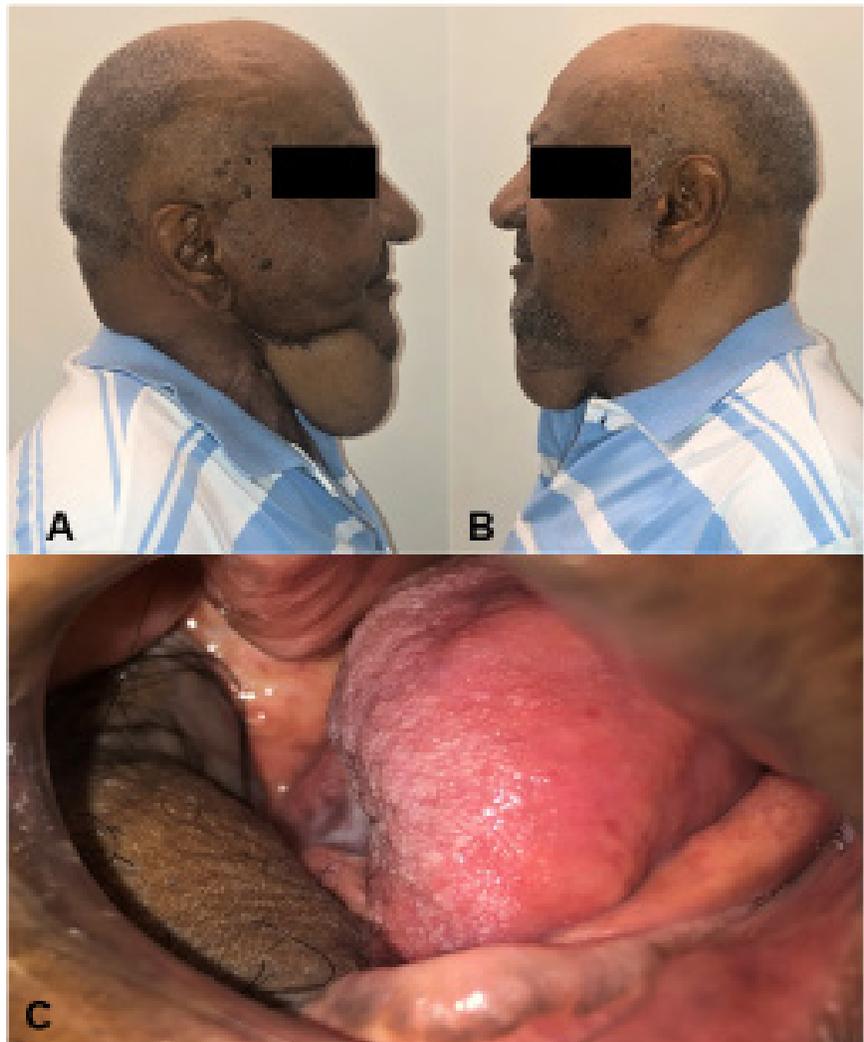
**Figure 1.** Oro-cutaneous fistulae (A), necrotic bone exposed in the right hemimandible (B), and orthopantomography, which evidenced osseous rarefaction regions simulating sequestra areas (C).

clinic with active and mucopurulent drainage in the initial right submandibular orocutaneous fistula region. Intraoral clinical examination showed a 2-cm long suppurative bone exposure with the presence of bone remnants and dehiscence of the initial surgical wound. With a clinical and radiographic diagnosis of refractory ORN, right hemimandibulectomy and surgical lavage were defined as therapeutic protocol and, upon referral to the Head and Neck Department, microsurgical reconstruction with musculocutaneous thigh flap was the conduct of choice. Submandibular access was done from the right, followed by subplatysmal flap elevation and deep plane breakdown for full exposure of mandibular stumps (Figure 2A). The devitalized necrotic



**Figure 2.** Six months after the first surgery, a second resection was performed due to orocutaneous fistulae with pus discharge (A). The right hemimandible was resected and removed to diminish acute local infection caused by refractory ORN (B). Moreover, facial reconstruction with musculocutaneous free flap was performed (C, D).

bone was then removed, and the mandibular stump margins were resected using a reciprocating saw (Figure 2B). Finally, osteoplasty of the remaining bone margin was done until a healthy bone tissue was obtained, followed by microsurgical reconstruction after surgical field wash with abundant saline (Figure 2C). The surgical technique to obtain the flap consisted of anterolateral thigh flap dissection, including a section of the vast lateralis muscle. The proximal dissection of the vascular pedicle was done, followed by the corresponding proximal ligation of the pedicle. The flap was then positioned and fixated for the reconstruction of the intraoral defect and skin coverage of the right and submental submandibular region. Afterward, flap revascularization was done by end-to-end microsurgical anastomoses with 9-0 nylon in the upper thyroid artery and the thyrolingofacial trunk (TLF) on the left. Finally, extra-oral flap suture and cervicotomy closure were done, followed by primary closure of the donor area (Figure 2D). After 3 months of the surgery (postoperative) and follow-up by the Department of Speech Therapy, Head and Neck, and Stomatology, the patient showed stability of the surgical field and musculocutaneous flap, with no signs of recurrence, infection



**Figure 3.** Follow-up three months after reconstruction by musculocutaneous free flap placed in the right hemimandible region, without signals of clinical infection or tissue dehiscence nor lost continuity on the flap.

and/or exposure of remaining bone tissue. The possibility of rehabilitation of the region will be evaluated later, along with a reduction of the flap size for both functional and aesthetic improvement (Figure 3).

### Discussion

There are currently several types of treatment for ORN, from conservative drug and topical therapy to mutilating resections for pathological jaw fractures. Hyperbaric oxygen therapy<sup>3</sup> has become one of the supporting alternatives to conventional treatment before and after surgical procedures on the jaws. In the case of conservative treatment, the use of antioxidant and vitamin-E based medication aims at accelerating tissue healing; besides, both are strong

antifibrotic agents (PENTOCLO)<sup>4</sup>. Antibiotics are used for decreasing the bacterial load on the wound—not as a definitive treatment to cure the wound itself. As for surgical treatment, the greatest advances have taken place in reconstructive surgery with the development of musculocutaneous flaps and the use of microvascularized free flaps, which favors both restorations of jaw continuity and supply of blood supplement for non-irradiated surrounding tissue<sup>4</sup>.

Resection and reconstructive surgeries are indicated in patients who failed to respond to conservative treatment, such as in superficial bone debridement with hyperbaric oxygen therapy and who evolved to a pathological fracture of the affected mandible<sup>5</sup>. On the other hand, these latter therapies are effective when properly indicated and, above all, for large bone and soft tissue defects, which will have a high functional and psychosocial impact on patients suffering from complex, mutilating clinical conditions. Microsurgical free flaps are the first choice in the case of extensive bone defects with wide resection margins since a non-irradiated flap with good vascularization would promote good healing and the remnant bone tissue that has been peripherally involved by the ORN<sup>5</sup> would be viable.

In the case of patients with a decreased systemic condition and old age, besides an unfavorable prognosis of their lesion, or with a posterior defect in the mandible, reconstruction with an osteomyocutaneous free microsurgical flap might be contraindicated. However, musculocutaneous flaps, which involve mainly soft tissue, become a feasible alternative for an aesthetic and functional rehabilitation with favorable results for the patient<sup>5</sup>.

The choice of musculocutaneous thigh flap, as described in the literature<sup>5</sup> is a viable alternative with satisfactory clinical results regarding the extensive reconstructions in which there is a double reconstruction to both restores remaining bone tissue and surrounding soft tissue. In the case report presented here, and due to the general and local condition of the patient, the first choice was the free thigh flap. After 3 months of follow-up, the patient presented a stable condition with the possibility of future rehabilitation in the affected maxillofacial region, with no signs of infection, exposure of remnant bone tissue and presence of extraoral fistula, which, according to the existing literature, would be the main and most common complication after reconstruction with free microsurgical flap<sup>5</sup>.

The fundamental principle for clinical success in the treatment of refractory ORN or ORN at advanced stages is the complete removal of necrotic bone tissue and immediate reconstruction of the injury with free microsurgical flaps for primary closure of the surgical injury, aiming at possible future rehabilitation of the maxillofacial region to improve both functional and aesthetic capacity. Although the literature reports various modalities of reconstruction with musculocutaneous and osteomusculocutaneous flaps, in this case report, both for the characteristics of the patient and of the region to be reconstructed, the musculocutaneous thigh flap was a satisfactory choice for reconstruction after three months of clinical-surgical follow-up. Prospective studies and case series with considerable follow-up are needed for greater clarification regarding the survival of these flaps in mandibular reconstructions due to ORN, as well as regarding factors associated both to the patient and to the site of osteoradionecrosis.

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