

Surgical Approach of Mandible Osteoradionecrosis Caused by Idiopathic Fracture

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Abordagem Cirúrgica de Osteoradionecrose Mandibular Causada por Fratura Idiopática

Abordaje Quirúrgico de Osteoradionecrosis Mandibular Causada por Fractura Idiopática

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ABSTRACT

Introduction: Osteoradionecrosis of the jaws affects 1% to 6% of patients undergoing radiotherapy and is considered the most severe oral complication resulting from this therapeutic modality. **Case report:** This is a 65-year-old man diagnosed with squamous cell carcinoma in the left oral floor, treated with surgery and adjuvant radiotherapy. In the dental evaluation, no clinical or radiographic changes were observed. Two weeks after the radiotherapy, the patient reported severe pain in the mandible, with no report of trauma or fall. The panoramic radiograph showed a fracture in the left mandibular body, suggesting an idiopathic fracture while asleep. After ten days, there was intraoral bone exposure of the distal portion of the mandible and conservative treatment with analgesia, superficial osteotomy under local anesthesia and prophylactic antibiotic therapy was performed. The patient evolved with purulent secretion, extraoral fistula and elimination of bone sequestration, after five months, confirming the diagnosis of osteoradionecrosis. Thereby, 11 months after the fracture diagnosis, the patient underwent a surgical intervention with partial mandibulectomy of the distal portion. After seven months of post-surgical follow-up, the patient presents neither clinical or radiographic evidence of osteoradionecrosis. **Conclusion:** The treatment of osteoradionecrosis is considered challenging for dentists who deal with this side effect of radiotherapy. Therefore, the importance of training the dentist to work in all stages of cancer treatment is highlighted.

Key words: Osteoradionecrosis/radiotherapy; Osteoradionecrosis/therapy; Mouth Neoplasms/complications; Mandibular Osteotomy; Medical Oncology.

RESUMO

Introdução: A osteoradionecrose acomete de 1% a 6% dos pacientes submetidos à radioterapia e é considerada a complicação oral mais grave advinda dessa modalidade terapêutica. **Relato do caso:** Trata-se de um homem, 65 anos, com diagnóstico de carcinoma de células escamosas em assoalho bucal esquerdo, tratado com cirurgia e radioterapia adjuvante. Na avaliação odontológica inicial, não foram observadas alterações clínicas ou radiográficas. Duas semanas após o término da radioterapia, o paciente relatou ter acordado com dor intensa em mandíbula, sem relato de trauma ou queda. A radiografia panorâmica evidenciou fratura no corpo mandibular esquerdo, sugerindo fratura idiopática durante o sono. Após dez dias, houve exposição óssea intraoral do coto distal e preconizou-se tratamento conservador com analgesia, osteotomia superficial sob anestesia local e antibioticoterapia profilática. O paciente evoluiu com secreção purulenta, fistula extraoral e eliminação de sequestro ósseo, após cinco meses, confirmando o diagnóstico de osteoradionecrose. Diante desse quadro, após 11 meses do diagnóstico da fratura, optou-se pela intervenção cirúrgica de mandibulectomia redutora de coto distal. Depois de sete meses de acompanhamento pós-cirúrgico, o paciente encontra-se sem evidências clínicas e radiográficas de osteoradionecrose. **Conclusão:** O tratamento da osteoradionecrose é considerado desafiador para os dentistas que lidam com essa sequela da radioterapia. Portanto, destaca-se a importância da capacitação do dentista para atuar em todas as etapas do tratamento oncológico.

Palavras-chave: Osteoradionecrose/radioterapia; Osteoradionecrose/terapia; Neoplasias Bucais/complicações; Osteotomia Mandibular; Oncologia.

RESUMEN

Introducción: La osteoradionecrosis afecta del 1% al 6% de los pacientes sometidos a radioterapia y se considera la complicación oral más grave resultante de esta modalidad terapéutica. **Relato del caso:** Hombre, 65 años, diagnosticado con carcinoma de células escamosas en el suelo de boca izquierdo, tratado con cirugía y radioterapia adyuvante. La evaluación odontológica no presentó alteraciones clínicas o radiográficas. Dos semanas después de concluir la radioterapia, él informó haber despertado con un fuerte dolor en la mandíbula, sin historia de trauma o caída. La radiografía panorámica mostró una fractura en el cuerpo mandibular izquierdo, lo que sugirió una fractura idiopática durante el sueño. Diez días después, hubo exposición ósea intraoral del muñón distal y fue empleado tratamiento conservador con analgesia, osteotomía superficial bajo anestesia local y antibiótico profiláctico. El paciente evolucionó con secreción purulenta, fistula extraoral y eliminación de sequestro óseo, pasados cinco meses, confirmando el diagnóstico de osteoradionecrosis. Así, pasados 11 meses del diagnóstico de fractura, fue indicada intervención quirúrgica de mandibulectomía reductora del muñón distal. Después de siete meses de la cirugía, no hay evidencias clínicas o radiográficas de osteoradionecrosis. **Conclusión:** El tratamiento de la osteoradionecrosis se considera un desafío para los dentistas que se ocupan de esta secuela de la radioterapia. Por lo tanto, se destaca la importancia de capacitar al dentista, para que trabaje en todas las etapas del tratamiento oncológico.

Palabras clave: Osteoradionecrosis/radioterapia; Osteoradionecrosis/terapia; Neoplasias de la Boca/complicaciones; Osteotomía Mandibular; Oncología Médica.

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INTRODUCTION

Osteoradionecrosis (ORN) affects 1% to 6% of patients undergoing radiotherapy and is considered the most severe oral complication resulting from this therapeutic modality since it can lead to devastating esthetic and functional consequences to the patient affected¹⁻⁴. It develops mainly in men's mandibles, older than 50 years, between four months and three years after the end of radiotherapy.

Conceptually, it is defined as an "area of exposed necrotic bone tissue that does not heal within a minimal period from three to six months in the absence of local neoplastic disease"^{3,7}. Its etiology is associated to risk factors such as doses above 66Gy, brachytherapy, size and location of the tumor, patient's age and comorbidities^{5,8,9}. The physiopathology is still inaccurate although a number of theories have been proposed^{10,11}.

The treatments proposed consist in prescription of antibiotics, anti-inflammatory, hyperbaric oxygen-therapy, ultrasound, and pentoxifylline associated to tocopherol (vitamin E) and clodronate. Also, surgical bone removal and recovered vascularized bone tissue transplantation can be performed^{4,5,12,13}. ORN management continues to be a challenge for dental surgeons working in this area¹⁰. Therefore, the objective of this study is to report a case of ORN resulting from mandibular idiopathic fracture showing the diagnostic process and therapeutic approach.

CASE REPORT

Retrospective case report of a 65 years old male patient with diagnosis of squamous cell carcinoma (SCC) in the left floor of the mouth. He presented the habit of smoking (rope) for 53 years, associated with alcohol consumption (one bottle of distilled beverage per day) for 47 years.

The physical exam revealed the presence of ulcer in the left floor of the mouth, extending to the lower alveolar ridge measuring 2.5 x 2.5 cm. Complementary, panoramic radiography was performed and no bone alterations were detected (Figure 1A).

Tumor staging was T2N0M0, stage II. The surgical treatment consisted in the pelvemandibulectomy of the left marginal horizontal ramus, left supraomohyoid cervical emptying and primary synthesis in oral cavity with sternocleidomastoid muscle flap. The surgical histopathological report showed moderately differentiated epidermoid carcinoma, infiltrating muscle tissue with perineural invasion and metastatic epidermoid carcinoma in level I left lymph node, staging pT1N1, stage III. Furthermore, adjuvant radiotherapy (61Gy in 33

fractions) utilizing the modality Cobalt 60 was performed, in the equipment Theratron-780C, 45 days after the surgery (Figure 1B).



Figure 1. A. Panoramic radiography prior to radiotherapy; B. Distribution of the dose in axial plan of the radiotherapy planning, equipment Theratron-780C, in software Eclipse, of Varian.

Fifteen days after the end of radiotherapy, the patient reported having awakened with intense pain in the left mandible with no episodes of recent trauma or fall. The intraoral exam showed no alteration, except for the spontaneous local pain, though the panoramic radiography revealed a fracture of the left mandibular body, suggesting diagnosis of idiopathic fracture while asleep (Figure 2A). After ten days, intraoral bone exposure was evidenced (Figure 2B and 2C) and initially, conservative conduct was recommended with outpatient follow up, orientations of oral hygiene, chlorhexidine 0.12% mouthwash followed by a superficial osteotomy under local anesthetic with prophylactic antibiotic therapy. After seven days, the clinical condition was unchanged.

Five months later, chlorhexidine 2% gel was also prescribed because of the accumulation of bacterial plaque over the exposed bone. The patient evolved with extraoral fistula and purulent exudate, being initiated antibiotic therapy and defined the diagnosis of ORN at advanced stage. The patient evolved with elimination of the bone sequestration through extraoral fistula associated with improvement of the clinical condition.

Eleven months after the fracture, the patient evolved with pain and limitation of mouth opening and through panoramic radiography, it was shown the rotation of the distal stump of the mandible (Figure 2D). Then, the surgical team opted for marginal mandibulectomy in this region. The histopathological analysis revealed fragments of neoplasm-free bone tissue and jugal mucosa showing epithelial hyperplasia without atypia. Seven months after the surgery, the patient was in good clinical condition, without clinical and radiography alterations (Figure 3).

The Institutional Review Board approved the study number CAAE 14282619.8.0000.5274 and report number 3,347,744, the patient read, agreed and signed the Informed Consent Form.

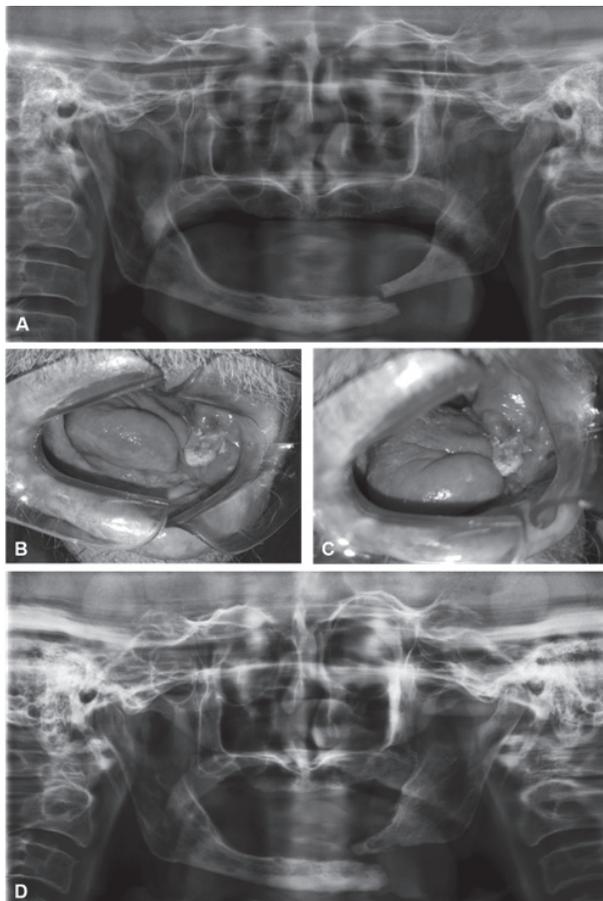


Figure 2. A. Panoramic radiography two weeks after radiotherapy showing mandibular fracture; **B and C.** Intraoral photos of the exposure of left mandible distal bone stump ten days after the diagnosis of the fracture; **D.** Panoramic radiography after 11 months of the diagnosis of mandibular fracture.

DISCUSSION

The incidence of ORN in the mandible is sevenfold higher when compared to the maxilla¹⁴, overall because is a more cortical, dense and less vascularized bone¹⁵. ORN is considered time-dependent and can occur at any moment after the end of radiotherapy¹⁶. The current study corroborates with the literature in terms of location and time of onset of ORN.

In the present case, some risk factors already evidenced by other authors were observed such as doses above 60 Gy, proximity of the tumor in relation to the bone, age of the patient and possible trauma of the mandible while asleep, causing fracture of the mandibular body^{5,8,9,15}. According to Owosho et al.¹⁷, doses above 60 Gy, even with the technique of intensity modulated radiation therapy (IMRT) are statistically associated with the increase of the risk of development of ORN¹⁷. Furthermore, marginal mandibulectomy to which the patient underwent is a procedure that aims to ensure neoplasm free-surgical margins, however, it can grant a critical vertical bone

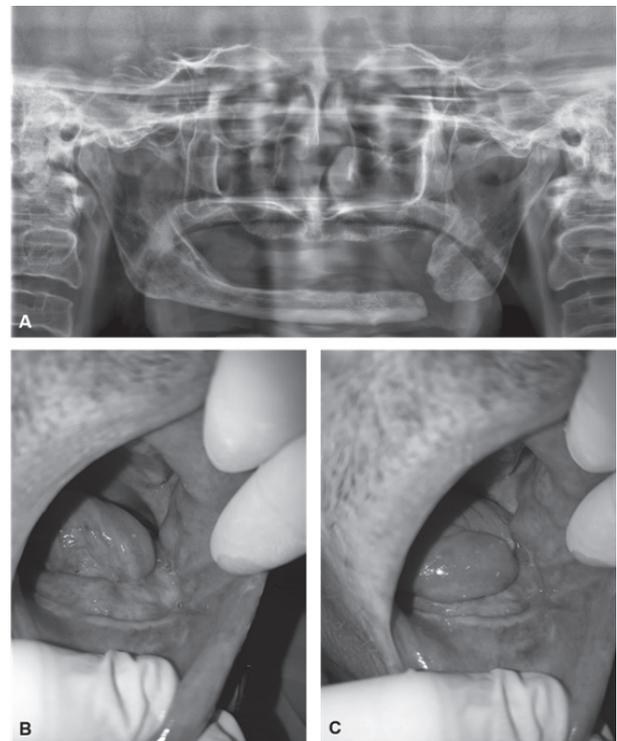


Figure 3. A. Panoramic radiography after seven months of marginal mandibulectomy; **B and C.** Intraoral photography of the region of the fracture after seven months of the surgery.

diameter, making the remained bone segment more susceptible to fracture, mainly during chewing¹⁸.

The fracture in the current clinical case occurred previously to ORN and the patient evolved with pain, trismus, formation of fistula, suppuration, elimination of bone sequestration and local infection, symptoms already reported in ORN conditions^{10,19}.

Despite the advances in the knowledge of ORN, its etiology remains unclarified. Initially, there was suspicion that it was caused by infection associated with difficulty of regeneration of tissues irradiated, resulting in bone necrosis²⁰. In 1983, Marx proposed the theory that the radiation leads to hypoxia, hypovascularization and tissue hypocellularity which eventually ended in necrosis and formation of the chronic disease as a wound that did not heal¹¹. Recently, the theory of the radiation inducing fibrosis was proposed, in which activation, deregulation of the proliferation and the metabolism of the fibroblasts would occur, creating an atrophic tissue^{1,10,21}.

For small bone exposures, conservative treatment is recommended only with drugs prescription^{4,5,12,13}, while in the severer cases, the option is surgical procedures²², as in the present case. The surgery must remove the necrotic tissue with safety margins until finding the vital bleeding bone and reconstruct the defect with vascularized soft tissue¹⁵. Chen et al.²³ observed that, after the surgical intervention associated or not to hyperbaric oxygenation,

the patients (98 of 105; 93.3%) completely healed from ORN²³.

Because of the uncertainties related to its etiology, ORN treatment continues to be challenging¹⁵ and demands the multi-disciplinary team to act, requiring the presence of the dental surgeon through all the stages, from prevention to the treatment of the oral toxicities and management of possible complications as ORN²⁵.

CONCLUSION

The management of ORN is challenging for dentists who deal with patients treated with radiotherapy of head and neck. The importance of multi-disciplinary team to manage this late sequela from radiotherapy is emphasized in view of the success of the treatment. The performance of the oncologic dental surgeon is ratified through all the stages, since the early diagnosis until the proper treatment.

CONTRIBUTIONS

Lísia Daltro Borges Alves, Ana Carolina dos Santos Menezes, Fernanda Vieira Heimlich, Débora Lima Pereira contributed substantially for the conception and/or design of the study. Marco Túlio Cunha Santos, Fernando Luiz Dias, Marcos Borges Moreto and José Roberto de Menezes Pontes contributed to obtain, analyze and/or interpretation of the data. Lísia Daltro Borges Alves, Débora Lima Pereira, Marco Túlio Cunha Santos, Héilton Spíndola Antunes participated of the wording and critical review. All the authors approved the final version to be published.

DECLARATION OF CONFLICT OF INTERESTS

There is no conflict of interests to declare.

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