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Advanced Osteoradionecrosis In The Maxilla With Pathological Fracture (Clinical Finding) Case Report

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ABSTRACT
Justification: Osteoradionecrosis of the jaw (ORN) is a known complication related to radiotherapy, having a great impact and may affect individuals undergoing radiotherapy for the treatment of head and neck cancer, and may be associated with an unfavorable dental condition. **Objective:** to report a clinical case of pathological fracture of the maxilla, as a consequence of osteoradionecrosis. **Conclusion:** Head and neck tumors represent approximately 10% of malignant tumors, 40% of which are in the oral cavity. The National Cancer Institute (Inca) forecast for Brazil, in 2023, is that 39,550 new cases of head and neck cancer will appear, including cancers of the oral cavity, thyroid and larynx. The need and importance of the dental surgeon inserted in a multidisciplinary team within an oncological team, has a preponderant role in the early diagnosis of neoplasms of the oral cavity and face; as well as in the treatment prior to radiotherapy and in maintaining the quality of life of this patient after treatment. Keywords: Oral cancer; Osteoradionecrosis; Pathological Fracture.

I. INTRODUCTION

Osteoradionecrosis was first described by C. Regaud in 1922, being one of the most severe oral complications of oncological treatment resulting from radiotherapy with high doses of irradiation in the orofacial region. It can occur between four and 24 months after starting treatment. The bone is exposed to the oral cavity, without vitality and without adequate healing for a period of three to six months. This condition is a combination of four factors: high radiation rate, hypoxia, hypocellularization and hypovascularization, causing bone necrosis, associated with a local traumatic or infectious factor.[1,2]

Osteoradionecrosis manifests itself most commonly in the posterior region of the mandible. The diagnosis of symptomatic complications is based on clinical characteristics, involving bone exposure in the oral and maxillofacial region, pathological fracture, infection, halitosis and formation of a fistula to the skin or oral mucosa. In asymptomatic complications, diagnosis can be made through imaging tests.[1,2]

Generally, osteoradionecrosis is associated with some signs and symptoms such as pain, trismus, chewing difficulties, extraoral or intraoral fistulas, pathological fracture, local infection and drainage of purulent secretion. Symptoms such as bad breath, paresthesia along the inferior alveolar nerve and dysesthesia. He also adds that initial patients have no symptoms, consequently diagnosis is delayed.[3,4]

Among oral cavity tumors treated with radiotherapy, osteoradionecrosis affects the mandible in 90% of cases, followed by the maxilla. Other bones of the face, such as the frontal and zygoma, are rarely affected. Historically, osteoradionecrosis of the jaw had an incidence rate of 2% to 22% of cases treated with radiotherapy. However, these rates have been declining, currently varying at around 5%, due to the advent of new radiotherapy techniques and preventive oral hygiene care.[5,6]

Radiotherapy as primary, adjuvant or palliative therapy is used in approximately 75% of patients with head and neck cancer, which significantly improves therapeutic efficiency.[7,8,9,10]

Every year, more than 8 million new cases of cancer occur worldwide, of which approximately

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900,000 are cases of malignant neoplasms of the upper aerodigestive tract. Head and neck tumors represent approximately 10% of malignant tumors, 40% of which are in the oral cavity, 25% in the larynx, 15% in the pharynx, 7% in the salivary glands and 13% elsewhere.[11,12,13,14]

The National Cancer Institute (Inca) forecast for Brazil, in 2023, is that 39,550 new cases of head and neck cancer will appear, including cancers of the oral cavity, thyroid and larynx. If we add melanoma skin cancer, which also affects the head and neck region, the number rises to 48,530. The regions of the country that lead the annual head and neck cancer estimates are the Southeast (20,470) and Northeast (10,070). They are followed by South (4,830), Central-West (2,760) and North (1,420). Bringing together all types of cancer, Inca estimates that 704,000 new cases will be diagnosed. This total is for each year of the three-year period 2023 – 2025, which means there will be 2.1 million in three years.[15,16]

The objective is to present a clinical case report, resulting from a clinical finding and imaging exams.

II. CASE DESCRIPTION

This is a clinical case report. Patient, male, J.A.I.; 66 years old. He was referred for an evaluation in Oral and Maxillofacial Surgery and Traumatology, a private clinic, for extraction of dental element 26. (Figure 1)

He had already undergone extraction of dental element 27. On intra-oral examination, he presented dysesthesia, mobility of the element, local infection, mucosal ulceration and bone exposure. He presented full mobility of the maxilla, corroborating the tomographic findings: thickening of the mucosa covering the maxillary sinuses, sinuous nasal septum, with deviation to the right side, absent left inferior nasal turbinate (suggestive of turbinectomy); in the region of the right upper nostril, hypodense images in soft tissue; a hypodense, osteolytic image is noted, with a "moth-eaten" appearance involving the posterior, lateral, medial, anterior walls and floor of both maxillary sinuses, hard palate and bilateral posterior maxilla extending to the region of teeth 15 and 23 , which in addition to teeth 24, 25, 26 are also involved in the injury. Imaging causes destruction of the cortices in these regions, with an increase in soft tissues. Image suggestive of odontogenic tumor, osteomyelitis or osteonecrosis. (Figures 2, 3, 4 and 5).

Fig. 1. Initial photo. Source: the author.

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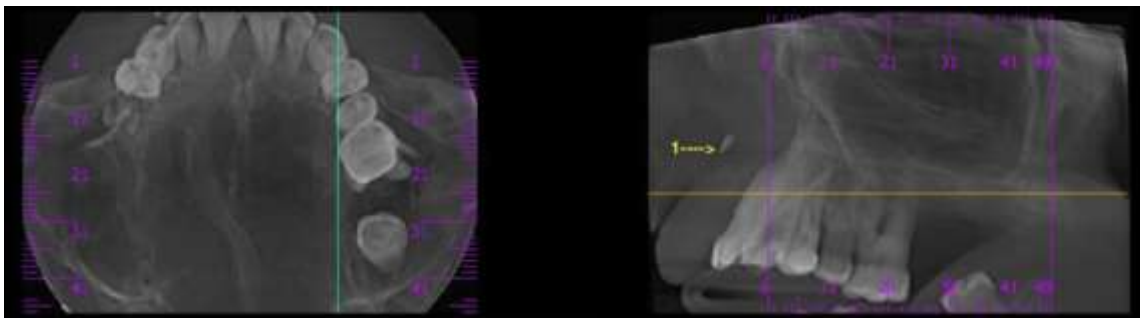

Fig. 2. Corresponding axial and sagittal sections. Source: the author.

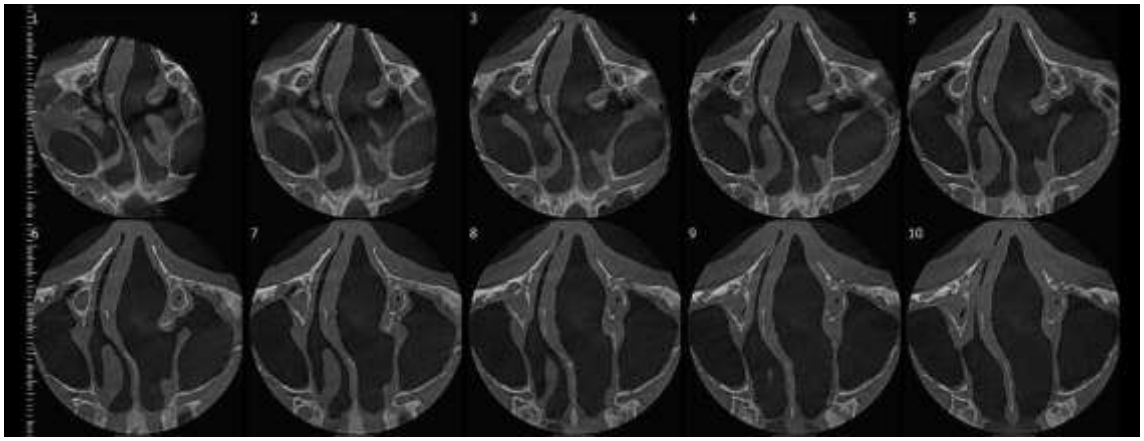

Fig. 3.Axial sections, showing the sinuous septal deviation to the right. Source: the author.

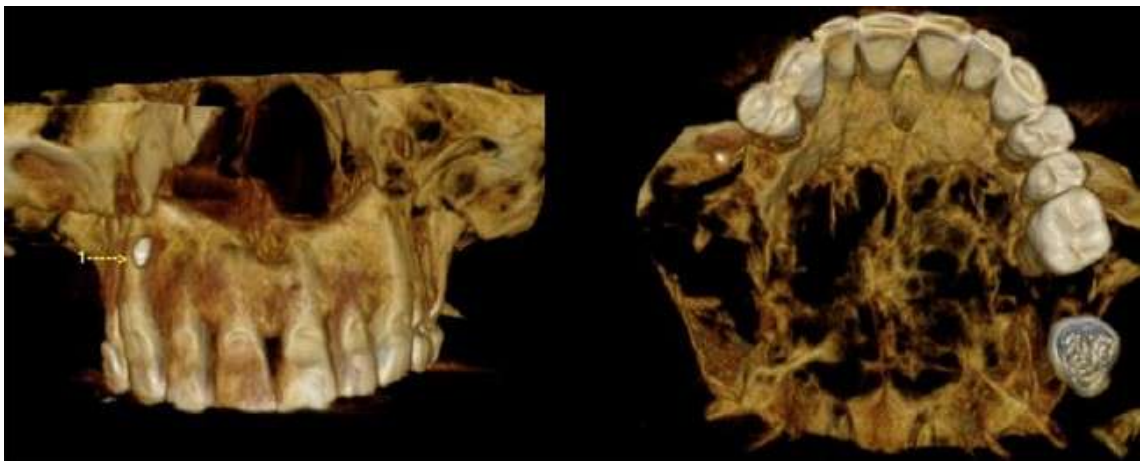

Fig. 4. 3d reconstruction. Hypodense, osteolytic image, with a “moth-eaten” appearance.

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Fig. 5. 3d reconstruction. Hypodense, osteolytic images, with a "moth-eaten" appearance and showing the pathological bilateral maxillary fracture line.

In 2017, he was diagnosed with maxillary sinus carcinoma, stage T2 (tumor limited to the antral mucosa of the suprastructure, without bone erosion or to the infrastructure with destruction of the medial or inferior bone walls. At the time, as treatment, it was underwent head and neck radiotherapy and chemotherapy and was unable to provide other information.

Maxillary sinus carcinoma is uncommon and represents 0.2%-0.8% of neoplasms, 3% of head and neck carcinomas and 80% of paranasal sinus tumors.[17,18,19]

The majority of tumors that occur in the maxillary antrum are of epithelial origin and squamous cell carcinomas account for more than 80% of all cases of malignant neoplasms, with adenocystic carcinoma being second in frequency. Most patients have advanced disease when symptoms begin. Computed tomography (CT) and magnetic resonance imaging (MRI) are well-established and useful techniques for evaluating the extension of the tumor to adjacent areas.[19,20]

The clinical and tomographic findings led us to refer the patient again to the oncology and head and neck surgery service; A new biopsy and a new histopathological examination were performed, where the tumor recurrence was observed. The patient is undergoing new treatment and is also monitored by our team.

In the literature, the recurrence rate varied between 0% and 50%, with an average of 23.15%. For local recurrences, which have already received radiotherapy in the initial treatment, surgery can be performed. If the first treatment was surgery, radiotherapy may be given. Chemotherapy or targeted therapy can be administered with radiotherapy or alone to treat recurrence that cannot be controlled by radiotherapy or surgery.[21,22]

III. DISCUSSION

Patients undergoing radiation therapy alone or as part of head and neck cancer treatment are more likely to develop osteoradionecrosis. The patient must be monitored by a multidisciplinary team before, during and after RT, to minimize possible adverse effects such as mucositis, osteoradionecrosis of the jaw (ORN), oral infections, dental changes, salivary disorders, dysgeusia and tendency to bleeding.[23]

Among the complications related to radiotherapy for the treatment of HNC, ORN of the jaw is a known and high-impact complication, affecting around 12% of individuals undergoing radiotherapy for the treatment of oral cavity cancer associated with an unfavorable dental condition.[4,23]

Initially, it was believed that the three factors that contributed to the development of osteoradionecrosis were radiation, trauma and infection.[24]

This understanding of the pathophysiology of osteoradionecrosis has evolved over time. The initial basic model focused on bone destruction secondary to trauma and infection after radiation. Marx offered an initial challenge to this paradigm in 1983, suggesting that microorganisms only served as contaminants in ORN samples and not as a determining cause of ORN. Instead, he proposed a model in which radiation led to hypoxic, hypocellular, and hypovascular tissue causing collapse and chronic non-healing wounds.[5,24,25]

Models of radiation-induced fibroatrophic processes, characterized by early inflammation followed by fibrosis and remodeling resulting in terminal tissue necrosis with reactive oxygen species and TGF-b1 playing prominent roles.[25,26,27]

Due to new evidence, it is also known that osteoclasts suffer effects related to radiation, promoting a suppression of bone turnover, leading to changes in bone remodeling and tissue damage.

This osteoclast disorder appears at early stages of the disease, even in the absence of vascular abnormalities. To support this theory, some authors noted a high incidence of jaw injuries similar to osteoradionecrosis in patients using bisphosphonate medications.[5,29,30]

According to the literature, the changes in the clinical signs of ORN that are most discussed remain the same; which consist, in most cases, of an area of exposed bone or a fistula that reaches the bone, tooth mobility or spontaneous tooth exfoliation. In imaging studies, they can vary from localized osteolytic areas, extensive osteolytic areas, sequestration and fracture. While the most common symptoms are pain and, in more severe cases, dysesthesia or anesthesia in the distribution of the inferior alveolar nerve in the mandible.[4,5,24,25,31]

The relevance of diagnostic agreement, within the clinical prevalence, where the affected site is within the radiation field of the head and neck; bone exposure; necrotic overlying bone; persistence of bone exposure for at least 3 months, with no tumor/recurrent metastases in the affected site.[32,33]

ORN is defined as loss of mucosal coverage and bone exposure lasting 3 to 6 months.[4,14,15,23,34] When ORN develops, patients suffer from severe pain, trismus, and deterioration in aesthetics such as cutaneous fistula oral. This further delays the rehabilitation of patients who have completed long-term cancer treatment. Furthermore, mandibular ORN often results in pathological fractures as it progresses.[34]

Patients with pathological fractures are required to undergo surgery. The most reliable treatment is resection and concomitant reconstruction. Surgery carries the risk of further dysfunction and aesthetic incapacity, and the stress for patients after severe cancer treatment is immeasurable. To control ORN infection and prevent pathological jaw fractures, understanding the occurrence is mandatory.[34,35]

The most common indication for aggressive surgery is a pathologic fracture. Therefore, we aimed to identify the risk factors for pathological fracture in patients with mandibular ORN. The present study included a long-term observational period (median, 79.0 months) and revealed that the incidence of pathological fracture in patients with mandibular ORN was 25.7%. The cumulative incidence rates of pathological fracture in patients with mandibular ORN at 36, 48 and 72 months after RT were 4.3%, 10.4% and 14.1%, respectively, increasing over time. Multivariate analysis showed that pathological fracture was associated with a greater number of mandibular fractures to teeth with a poor prognosis when the fracture occurred.[34,35]

Such studies have focused on the role of infection in exacerbating ORN. Infection control is an important factor in the treatment of ORN. Our hypothesis is that the accumulation of infection is an exacerbating factor for ORN and may be a trigger for pathological fracture, with the infection rate of patients with fracture being higher (73.7% vs 56.4%; P=0.276). However, further investigations are necessary to elucidate the role of bacterial infection in the occurrence and exacerbation of ORN.[33,34,35]

IV. CONCLUSION

As in our case, the tumor data were basic; doses and intervals of radiotherapy taken not reported; Due to the severity of the damage and the recurrence of the tumor, we decided to refer the patient to the head and neck surgery and oncology service.

Osteoradionecrosis of the jaws after radiotherapy for head and neck cancer represents an alternative diagnostic and therapeutic challenge for which multidisciplinary collaboration is essential.

In contrast to a mandibular predominance, maxillary ORN has not been widely publicized, due to the incidence and prevalence of ORN in the maxilla being rare; making this case one of the few clinically reported. The literature studied showed that resection and reconstruction methods should be selected after careful assessment of the state of soft tissues, extent of dead bone and the physical condition of the patients.

Dentistry must be essential for patients diagnosed with neoplasms, as well as chemotherapy, radiotherapy or individuals undergoing any other cancer treatment methods. In this segment, the dental surgeon works to minimize the discomfort caused during this process, provides an improvement in the quality of life, acts in early diagnosis, and provides adequate management regarding the particularities of this patient.

Finally, the need for dentistry linked to a multidisciplinary oncology team is identified, which in turn requires careful and individualized management for each type of treatment choice, patient age, systemic conditions and disease evolution. Thus, this will be assisted in a holistic way, providing a better quality of life and comfort for the patient.

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