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Osteonecrosis of the Jaw in a Patient with Multiple Myeloma: A Case Report

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Abstract

This clinical case describes a 60-year-old man diagnosed with multiple myeloma (MM), treated with chemotherapy and bisphosphonates that later developed osteonecrosis of the mandible. Histopathological examination of the lesion made it possible to associate bisphosphonate therapy as the cause of the pathology, thus ruling out neoplastic involvement.

Keywords: Bisphosphonates; Multiple Myeloma; Osteonecrosis of The Jaw; Oncology; Metastasis

Introduction

Multiple myeloma is a haematological neoplasm that can manifest with unique involvement of the mandible [1]. One of the therapies used to prevent the occurrence of skeletal lesions in MM is bisphosphonates, due to their role in inhibiting osteoclasts [2]. but it is known that they can cause osteonecrosis. This clinical case reports the occurrence of osteonecrosis of the mandible in a patient with multiple myeloma undergoing treatment with bisphosphonates. The challenge in this case was to identify the etiology of the osteonecrosis, which could be a bone metastasis from the MM or an adverse effect of the associated therapy. Histological examination is the key to the diagnosis.

Clinical Case Presentation

We present the case of a 60-year-old man, diagnosed with MM in May 2022, who underwent 4 cycles of chemotherapy (Velcade (bortezomib) + Revlimid (lenalidomide) + dexamethasone (VRd))- from May to September 2022 - combined with bisphosphonates (Zolendronic Acid) - from June to August 2022.

In October 2022, he came to the dental clinic referring intense and continuous pain in the right posterior mandibular region.

Objective examination through the vestibular area revealed gingival edema, suppuration involving teeth 45 to 48 and grade 3 mobility, according to Miller's classification, on teeth 46, 47 and 48 [3]. On the lingual side, it was possible to evaluate, moderate gingival inflammation and approximately 2.5mm of necrotic bone exposed on the external cortex (figure 1).

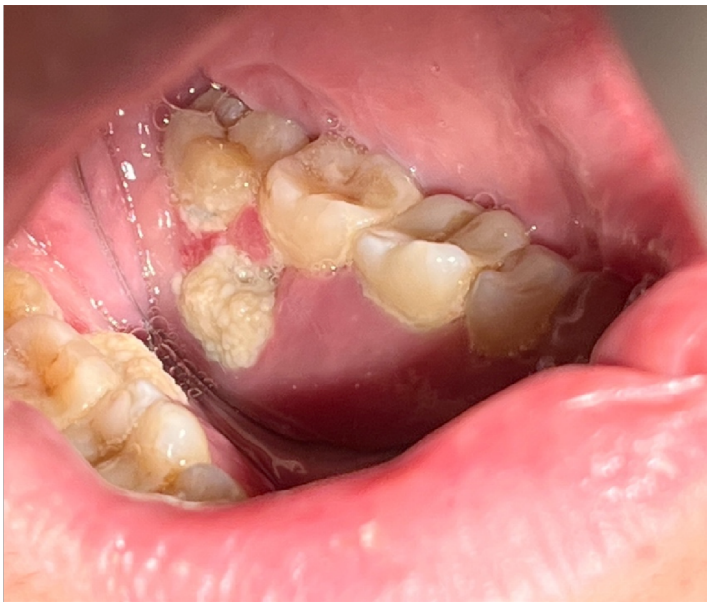

Figure 1: Lingual view of the necrotic bone.

Orthopantomography (figure 2) and periapical radiography (figure 3) were requested as complementary diagnostic exams. As neither revealed any findings, a cone beam computed tomography (CBCT) scan was requested (figure 3,4,6).

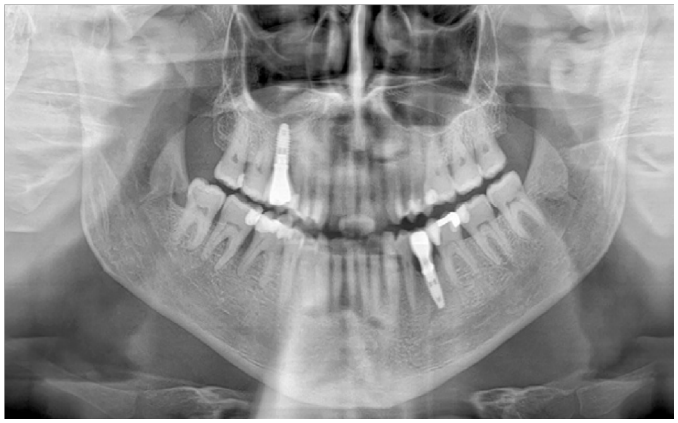

Figure 2: Initial panoramic x-ray

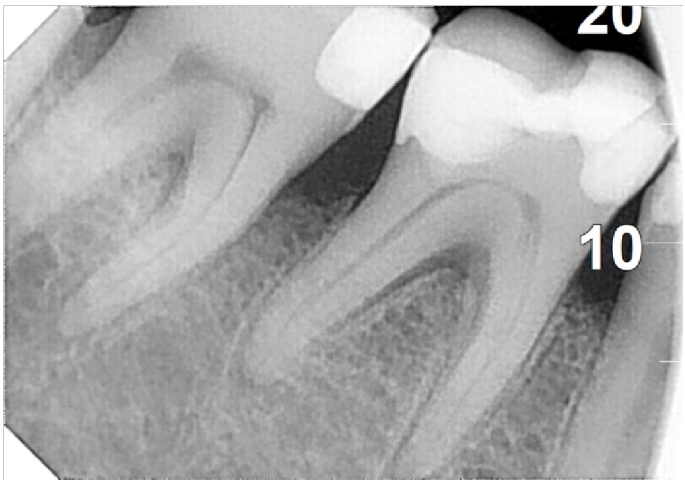

Figure 3: Periapical x-ray

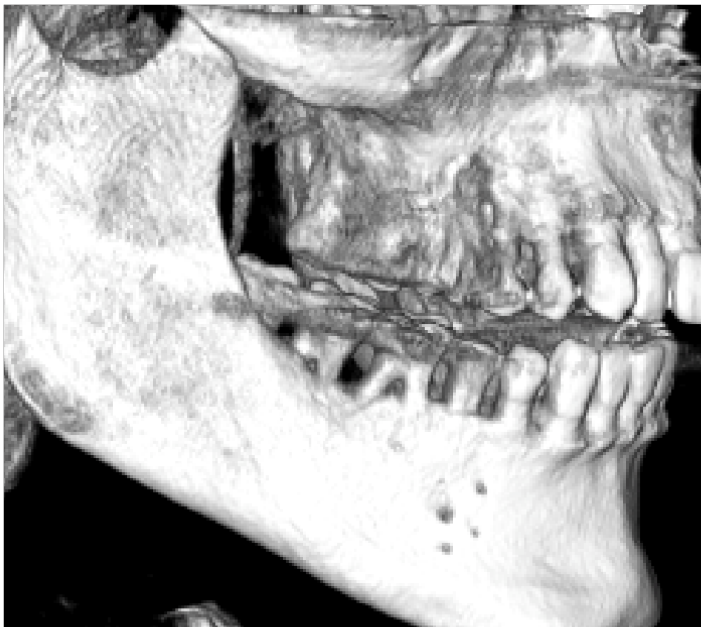

Figure 4: CBCT (buccal view)

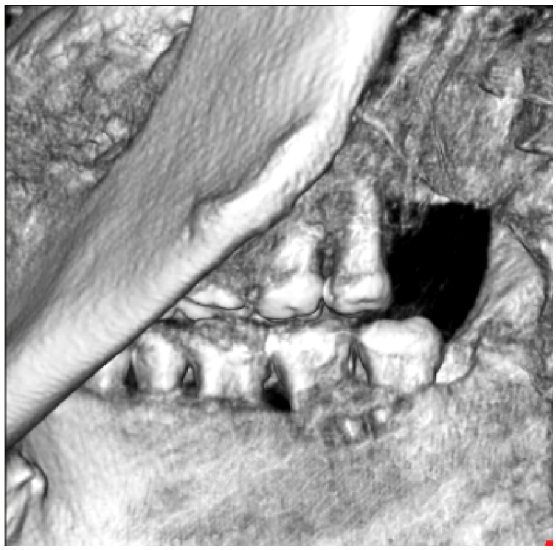

Figure 5: CBCT (lingual view)

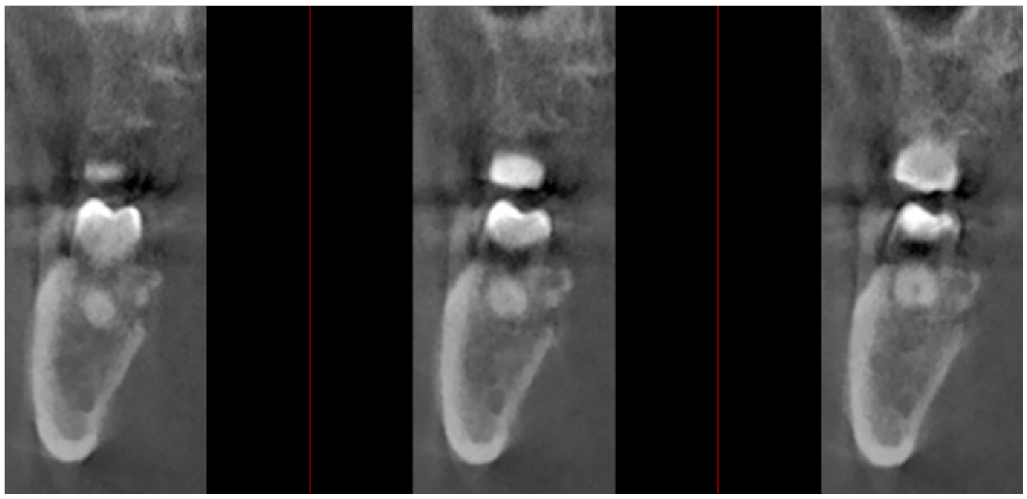

Figure 6: CBCT (axial view)

The first approach in this case involved local prophylaxis with an ultrasonic scaler and prescription of a non-steroidal anti-inflammatory (ibuprofen 600mg), 0.2% chlorohexidine mouthwash, and antibiotic therapy (amoxicillin-clavulanic acid 875-125mg).

The 7-day follow-up revealed significant improvements, maintaining pain only when chewing and improvement of periodontal insertion (mobility: tooth 45 - grade 1, teeth 46 and 48 - grade 2, tooth 47 - grade 3), showing stability in the extension of necrotic bone. In order to ensure masticatory comfort, a splint involving teeth 45, 46, 47, and 48 was made. At this stage, regarding the extension of the disease, a whole-body scintigraphy Tc99m-Medronate was carried out with a tomographic study that revealed a single, focal lesion with increased osteoblastic activity, located on the horizontal ramus of the right hemi-mandible (figure 7).

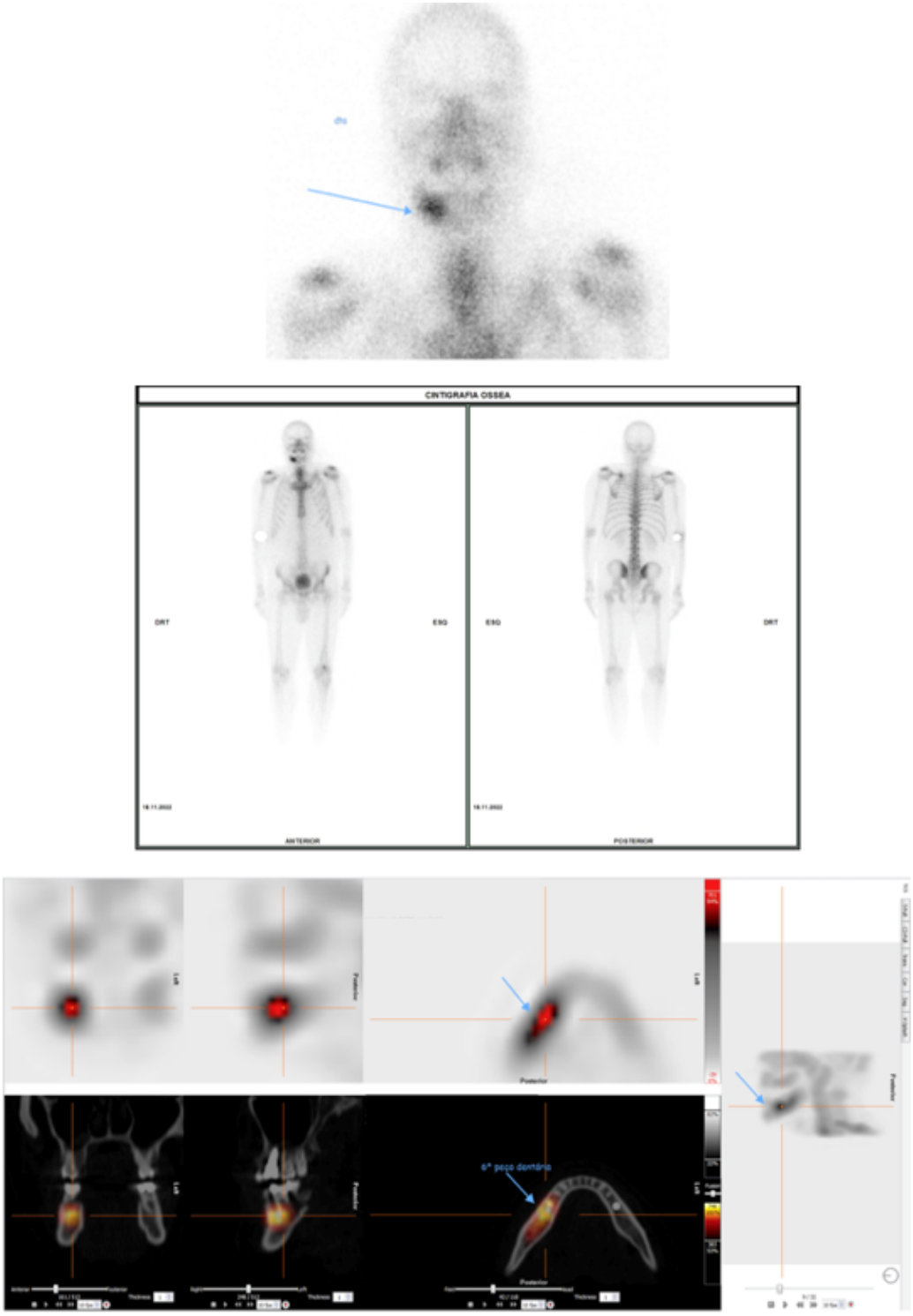

Figure 7: Bone scintigraphy

The third follow-up, 1 month after starting treatment, the amount of exposed bone was bigger, reaching approximately 4 mm of exposed necrotic bone (figure 8), still involving only the lingual plate.

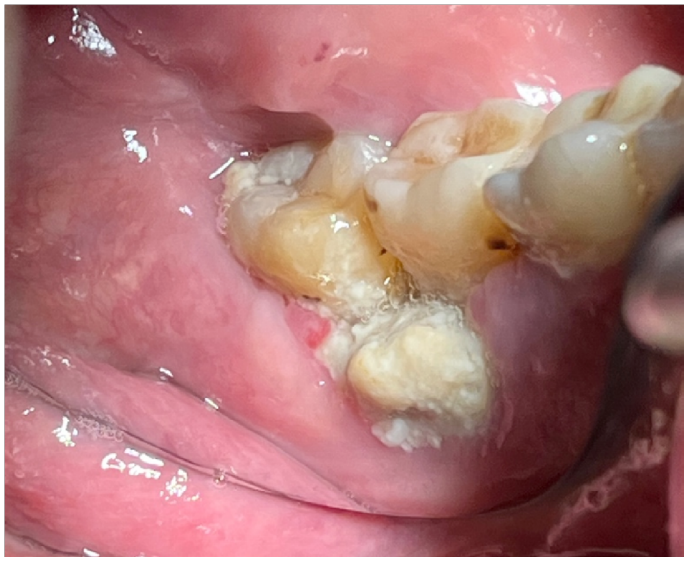

Figure 8: Third follow-up

After the multidisciplinary meeting, attended by the Oncology, Dental Medicine and Maxillofacial Surgery, all departments agreed that the surgery should be scheduled according to the 6-month safety window, given the therapy used to treat MM.

In January 2023, the maxillofacial surgery team carried out the surgery, removing all the affected bone and its margins (1.5 x 1.1 x 0.8cm) and extracting teeth 47 and 48.

Anatomopathological analysis revealed necrotic bone tissue and fibrinogranulocyte exudate, and colonies of gram-positive bacteria (Actinomyces).

In this way, it is possible to conclude the etiology of the necrosis, in which osteonecrosis caused by bisphosphonate therapy is emphasized, rather than a metastasis. We would like to point out the essential role played by the anatomopathological result in reaching a definitive diagnosis, making it possible to rule out oncological involvement.

The forth follow-up took place one month post-op, revealing complete healing of the soft tissues (figure 9) and healthy periodontal tissues. Tooth 45 is now under physiological mobility and tooth 46 shows a grade 1 mobility due to the lack of bone involving the distal root (figure 10). After this, patient was discharged and referred back to the oncology team in order to perform the autotransplant.

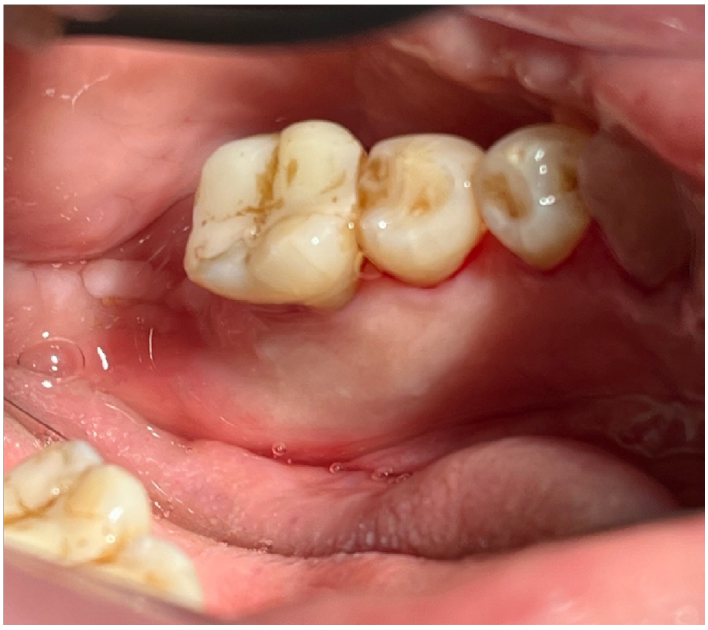

Figure 9: Forth follow-up

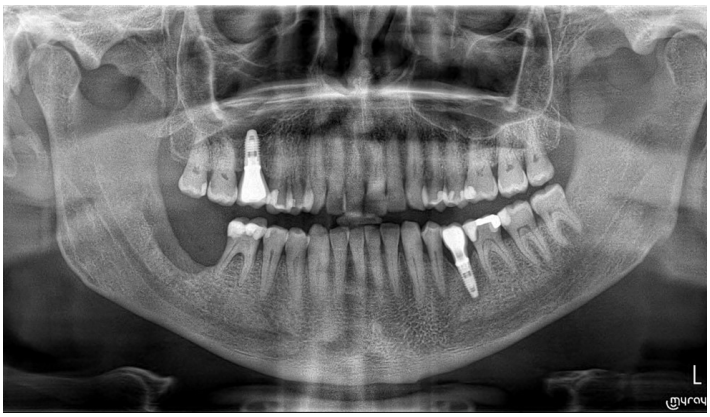

Figure 10: Panoramic x-ray (forth follow-up)

Discussion

The uniqueness of this clinical case is essentially based on two aspects: the first reflects the initial difficulty in understanding the etiology of the documented osteonecrosis, the second one is the ideal time to perform the surgical intervention in a patient who had recently taken bisphosphonates, so as not to aggravate the osteonecrosis that had already started. Regarding the etiology, the bone biopsy performed, which did not document plasmacytosis or monoclonality, in the only place where scintigraphy revealed bone proliferation, led to the conclusion of osteonecrosis associated with bisphosphonates. On the other hand, regarding the safety window

to perform the surgery, a period of 6 months was chosen, although the ideal period is not definitively established in the literature.

MM is a hematological neoplasm characterized by the progression of monoclonal plasma cells in the bone marrow, which can lead to lytic bone lesions, pathological fractures, and, for these reasons, pain [1,3], and can even occur in isolation in the mandible [1,4]. As far as the treatment of MM is concerned, in addition to chemotherapy and autotransplantation, bisphosphonates play an important role, in this case as a preventative measure [5], preventing bone lesions. This role is due to their mechanism of action by inhibiting the differentiation of osteoclasts, which reduces their activity and promotes apoptosis [2]. However, one of the complications associated with its use is osteonecrosis of the jaw [6,7], but in order to establish this causal relationship between the drug and osteonecrosis, it is essential to exclude osteonecrosis due to neoplastic disease [6]. It is also known that oral surgery carries an increased risk of developing osteonecrosis in patients taking (or having recently taken) bisphosphonates. For this reason, some authors [6,7] have tried to report, through a literature review, on the best regimen for continuing/suspending bisphosphonates in the treatment of dental lesions, in order to prevent osteonecrosis of the jaw, and concluded that is no clear benefit in discontinuing the drug. Kalra et al [2] recommend an individualized approach to the patient, as was done in the case described. Furthermore, osteonecrosis of the jaw in the context of bisphosphonates can mask the presence of MM [5,8], so special attention should be made to the assessment of these patients - in this case, only the biopsy performed made it possible to safely conclude that it was a bisphosphonate-related osteonecrosis of the jaw.

Conclusion

Differentiating between osteonecrosis of the jaw in the context of metastasis or associated with bisphosphonate medication is difficult, especially in a patient with a previous diagnosis of MM. However, although it is difficult, it is fundamental in orientating the patient, with implications for prognosis and appropriate treatment. In this case, the bone biopsy proved to be fundamental in clarifying the etiology of the necrotic lesion.

Additional Information

Human subjects: Consent was obtained by all participants in this study. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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