Mandibular lesion as the first sign of multiple myeloma in a young patient

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Abstract Multiple myeloma (MM) is a malignant neoplasm characterized by an abnormal intramedullary plasma cell proliferation. It accounts for approximately 10% of all hematologic malignancies. It generally occurs between fourth and seventh decades of life, and it is predominant in male patients. Patients lower than 40 years are considered young, and only very few cases have been reported. We present a rare case of a 27-year-old man whose initial findings of MM were a swelling at the ramus of the mandible.

Keywords: Mandible, multiple myeloma, oral cavity, plasma cell

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INTRODUCTION

Multiple myeloma (MM) is a malignant blood neoplasm characterized by an abnormal intramedullary proliferation of bone marrow cells and hypersecretion of monoclonal immunoglobulins.^[1,2] It represents approximately 10% of all hematologic malignancies.^[3] It generally occurs between fourth and seventh decades of life^[4] and is predominant in male patients.^[5,6] Patients diagnosed before the age of 40 are rare, and they are considered young in relation to this disease. Here, we report a 27-year-old Amazonian man whose initial findings of MM were a right mandibular ramus swelling.

CASE REPORT

A 27-year-old Brazilian–Amazonian man presented to the Surgery and Oral Pathology Service of the João de Barros Barreto University Hospital, Federal University of Pará,

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Belém, Pará, Brazil, with a complaint of a painful swelling in the right ramus of the mandible that had been presented for approximately 1 month. The medical history for the patient revealed no relevant contributory conditions. The clinical examination revealed facial asymmetry evidenced by an enlargement in the right posterior region of the mandible, and the patient also reported fatigue and weight loss. Intraorally, a discrete swelling in the buccal mucosa extended to the retromolar space [Figure 1a]. The panoramic radiograph revealed a multilocular ill-defined radiolucent lesion on the right side, expanding to the body, angle and ramus of the mandible [Figure 1b]. The computed tomography scan evidenced a hypodense area causing destruction of the vestibular and lingual bone cortical areas. Such area involved the body, angle and ramus of the mandible [Figure 1c]. The lesion's aspiration was negative. An incisional biopsy was performed under local anesthesia, and a tissue sample was removed from the

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interception zone of the ramus and body of the mandible. Results of microscopic analysis revealed fragments of malignancy presenting as compact sheets of atypical diffuse infiltrate of plasma cells. The neoplastic plasma cells presented as varied in size with eccentric nuclei, rounded and irregular formats. In some neoplastic cells, the nuclear chromatin presented as vesicle-patterned or delicate beads as well as prominent nucleoli [Figure 2a and b]. The immunohistochemical reactions were positive for CD138 [Figure 2c], plasma cell [Figure 2d], monoclonal to kappa



Figure 1: Clinical and image features of multiple myeloma of the reported case. (a) The presence of a discrete swelling in the buccal mucosa extended to the retromolar space. (b) A multilocular ill-defined radiolucent lesion in the right body of the mandible. Thinning of the right inferior cortical of the base and angle of the mandible was also evident. (c) A hypodense mass causing destruction of the vestibular and lingual bone cortical areas, with involvement of the body, angle and ramus of the mandible. (d) Bone scintigraph in the left seventh and tenth ribs, knees, heels and shoulders

[Figure 2e] and high Ki 67 immunostaining [Figure 2f] and were negative for leukocyte common antigen, desmin and citoqueratin. It was necessary to assess the possibility of involvement of other bones. The bone scintigraphy showed a mild radiopharmaceutical hyperconcentration in the left seventh and tenth ribs as well as in the knees and heels. In addition, a moderate hyperconcentration in the shoulders was noted [Figure 1d]. Thus, the diagnosis of MM was made, and the patient was referred to hematology and oncology department for treatment but eventually died 1 month after the diagnosis from pulmonary failure complications.

DISCUSSION

MM is a malignant neoplasm of plasma cells^[7,8] and is characterized by an abnormal proliferation of immunoglobulin-secreting plasma cells, which may produce M-protein, light-chain proteins (κ or λ) and cytokines.^[5,9] Plasma cell neoplasms are divided into three groups: MM, solitary plasmacytoma and extramedullary plasmacytoma.^[7,9] MM is more commonly found in patients between 40 and 70 years old,^[6] with a mean age of 60 years and males more affected,^[9] with the most affected location being the mandible. In the current case report, we did not consider MM as diagnosis hypothesis since it is more common in the elderly and rarely occurs in young patients.^[1-14]

On presentation of symptoms, tooth mobility, pain and soft-tissue swelling may occur in association with mandibular involvement.^[14] In this article, our patient complained of pain in the mandible, and a discrete



Figure 2: Microscopic features: (a) A histopathological examination of the specimen reveals neoplastic fragments of compact sheets of atypical diffuse infiltrate of plasma cells (H and E, ×100); (b) malignancy with plasma cells as varied in size with eccentric nuclei, rounded and irregular formats. In some neoplastic cells, the nuclear chromatin presented as vesicle-patterned or delicate beads as well as prominent nucleoli (H and E, ×200); (c) immunohistochemical staining showing positivity of cell to CD138 (×200); (d) plasma cell (×200); (e) monoclonal kappa (×200) and (f) high grades for Ki-67 (×200)

swelling on intraoral examination was observed, as earlier described. The radiographic characteristics of MM may vary from normal to diffuse osteosclerosis although multiple radiolucent bone lesions are the most common findings,^[10,12] which were observed in this presented case. Oral lesions rarely occur as the first sign of the disease,^[7,11] and few cases present oral lesions as the first sign of the abnormality, considering a systemic workup necessary to investigate the existence of systemic disease.^[9,11] Vertebrae, skull, pelvic bones, ribs, humerus and femur are commonly affected, in this order of frequency.^[5,13] In our case report, there was involvement of ribs, knees and heel bones, which was detected by bone scintigraphy and confirmed the final diagnosis.

Thus, we present a rare case of MM in mandible as the first sign of the disease in a young adult patient. It is important that oral surgeons and dentists bear in mind that oral lesions may be the first clinical sign of MM.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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