

Case Report

A multilocular radiolucency of mandible as the first evidence of multiple myeloma: A clinico-radiographic case report

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ABSTRACT

The incidence of multiple myeloma (MM) affecting the jaws is 30% and on rare occasions the oral involvement can be the first indication of the disease. Authors report a case of MM in a 40-year-old woman who presented with a multilocular radiolucent lesion in the left mandible initially mistaken as an ameloblastoma. Conventional radiographs revealed a multilocular lesion on the molar region. Computed tomography (CT) and 3 dimensional CT revealed lytic, space occupying lesion perforating the inferior cortex. Magnetic resonance imaging (MRI) revealed a hypointense lesion on T1 weighted image and hyperintense lesion on T2 weighted image. Histopathological and lab investigations lead to the diagnosis of MM. MRI is superior in depicting the size of the lesion as compared to CT and conventional radiographs.

Key Words: Magnetic resonance imaging, mandible, multilocular radiolucent lesion, multiple myeloma

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INTRODUCTION

Plasma cell dyscrasias may present as one of the three distinct clinical entities: Multiple myeloma (MM), solitary plasmacytoma of bone, and extramedullary plasmacytoma. MM accounts for 65% of plasma cell dyscrasias.^[1]

The cause of MM is unknown. Radiation exposure increases the risk as evidenced by a higher than expected rate of disease in atomic bomb survivors, radiation workers, and post irradiated patients with ankylosing spondylitis.^[2]

The most frequent presentation of MM is that of a disseminated disease with involvement of multiple skeletal sites. Because of its tendency for widespread

manifestations in multiple organs, this disease is of interest to many medical specialists, including dental surgeons.^[3] When MM occurs in mandible as a primary manifestation, it can pose a diagnostic problem as the presentation is similar to other cystic and odontogenic lesions.

A case of MM presenting as multilocular jaw tumor is discussed with a radiographic and biochemical work-up.

CASE REPORT

A 40-year-old woman reported to the dental out-patient department with a 4 month old painless progressive swelling in the left mandibular body. Swelling was first noticed by the patient when pain occurred on chewing sugarcane.

Physical examination showed facial asymmetry due to left lower facial swelling. Over the swelling the skin was of normal color, the facial musculature had normal tonicity and range of activity and the cranial nerve examination revealed no specific deficits. No regional lymphadenopathy was present.

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Intra-orally a firm to hard non-tender, non-pulsatile swelling measuring approximately, 3 cm × 1.5 cm on the left premolar-molar region, expanding the buccal and lingual plates was noted. Mandibular left second premolar, first molar, and second molar did not respond to vitalometry. Overlying mucosa showed no changes in consistency or color [Figure 1a].

Panoramic and occlusal radiographs revealed a multilocular radiolucent lesion of 3 cm × 2 cm, causing buccolingual cortical expansion [Figures 1b and c]. Periphery of the radiolucency was well-defined, but without a sclerotic reaction. The radiolucency presented, with thick internal septa, dividing it into five irregular locules [Figure 1b] with no evidence of trabaculae. The lesion caused root resorption of first and second molar.

Differential diagnosis of ameloblastoma, giant cell tumor, malignant jaw tumor, metastatic carcinoma, and odontogenic cyst was considered.

Pre-operative hematological analysis revealed normocytic normochromic anemia (6.50 g/dl), raised erythrocyte sedimentation rate (58 mm for 1st h), leucopenia (0.87 thousand/cu mm) with mild neutropenia (1.15 thousand per cu mm) and thrombocytopenia (138 thousand/cu mm). Macrocytosis, anisocytosis, and polychromatophils were seen on peripheral smear.

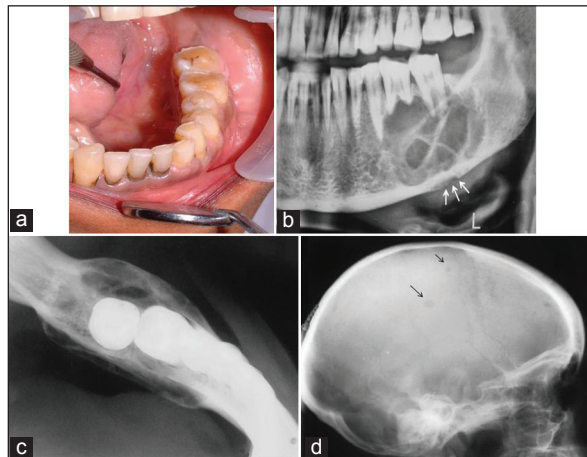


Figure 1: (a) Intra oral photograph showing mild expansion with obliteration of buccal vestibule. (b) Cropped panoramic view showing multilocular radiolucency with thick internal septae. Notice the destruction of the superior surface of the inferior mandibular cortex. (c) Left mandibular lateral occlusal radiograph showing minimal bucco-lingual cortical expansion due to the multilocular radiolucency. (d) Lateral skull radiograph showing multiple punched out lesions

An incisional biopsy revealed aggregates of plasma cells in loose connective tissue suggestive of MM.

Further tests showed hyperuricemia and hyperproteinemia with a decreased A:G ratio. A characteristic monoclonal gammopathy (“M” spike) was seen in the gamma globulin region on serum protein electrophoresis. Bone marrow biopsy demonstrated 90% infiltration by abnormal forms of plasma cells.

A complete radiographic survey, to rule out solitary plasmacytoma, revealed multiple punched out radiolucent lesions, on the lateral skull radiograph [Figure 1d] and two punched out lesions, on the right tibia.

Computed tomography (CT) revealed multiple lytic lesions on the inner and outer tables of skull, and a large, expansile, soft-tissue density, space occupying lesion, measuring 20 mm × 15 mm, on the left mandibular body [Figure 2a].

Magnetic resonance imaging (MRI) included sequences such as spin-echo (SE) (T1 weighted and T2 weighted), gradient echo (T2 weighted) and short tau inversion recovery (STIR). SE and fast SE

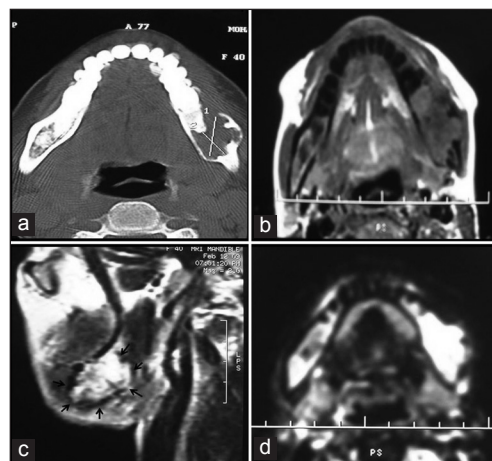


Figure 2: (a) Computed tomography bone window of mandible showing lytic, expansile, space occupying lesion of left mandibular body. Magnetic resonance imaging patterns of multiple myeloma manifesting as a mandibular lesion. (b) Axial T1-weighted spin-echo image showing focal lytic expansile mass with diffuse hypointensity compared to the hyperintensity of the background marrow. (c) Sagittal T2-weighted image revealing cellular marrow demonstrating reversal of normal magnetic resonance pattern, with diffuse hyperintensity of the lesion on a hypointense background marrow. (d) Axial short tau inversion recovery image showing no change in diffuse hyperintense signals of the lesion

revealed a large, solid, expansile, diffusely infiltrating space occupying lesion with ill-defined borders in the mandible. T1 weighted images showed focal hypointense, [Figure 2b] and T2 weighted images showed a hyperintense lesion [Figure 2c] suggestive of increased cellularity in the lesion, supporting the diagnosis. No change in signal intensity of the lesion, on fat suppression technique i.e., STIR in our case, was suggestive of diffuse involvement within the lesion [Figure 2d].

The patient was referred to an oncologist and is currently on chemotherapy with zoledronic acid-4 mg IV, thalidomide, and dexamethasone. A 4 month follow-up revealed no progression in the disease status.

DISCUSSION

MM is also known as Kahler's disease after Otto Kahler in 1889.^[4] MM is a clonal B lymphocyte neoplasm of terminally differentiated plasma cells.^[2] It occurs in the age group of 40-70 years, most commonly seen in males, with a M:F ratio being 4:1.^[5] In our case, the patient was a female of 40 years.

Approximately, 70% of MM produces, predominantly immunoglobulin's G (IgG), whereas 30% produce IgA, IgD or E secretory myelomas and non-secretory myelomas being rare. The light chains of IgG are kidney permeable and are detected as proteinuria or Bence-Jones proteinuria.^[3] In the present case, the tumor produced IgG, but Bence-Jones proteinuria was absent.

Up to 30% of cases manifest in the jaws.^[1] Primary lesion in the jaws accounts for 8% to 15% and that alone in mandible is reported to be 5.18%.^[6] In jaws, it manifests with pain, paresthesia, swelling, soft-tissue mass, mobility, tooth migration, hemorrhage, amyloid deposits in tongue, and pathological fractures.^[1,7] In our case, the first sign of this systemic malignancy was a mandibular swelling. Root resorption is not a frequent finding in MM. In the present case, root resorption in the mandibular left first and second molar can be explained by the encroachment from the developing osteolytic lesion.^[8]

In the jaws, the radiological findings reported are multiple punched out radiolucencies without sclerosing border, radiolucent lesions, osteoporotic and rarely osteosclerotic changes.^[1] The radiolucent mandibular lesion described in our case had, thick

septa dividing the radiolucent lesion into five irregular locules, defined borders, but without sclerotic reactions and erosion of the inferior cortex.

Staging of MM is carried out using the Salmon-Durie system based on conventional radiographs, CT, MRI, and nuclear imaging to differentiate patients between Stages I and III.^[9]

MRI is reported to be very sensitive in determining the exact location, size, and local compressive effects of focal plasmacytomas, the results of treatment, areas of potential complications, and the sites of focal disease for safe bone biopsies.^[2]

CT depicts the effect of lesion on bone, whereas MRI images the bone marrow directly. According to the literature, MRI is clearly superior to plain radiographs, but probably superior to CT too.^[2,9] In the present case, magnetic resonance was more precise, with the depiction of size and extent of the lesion in comparison to CT. The size of the lesion on MRI was 25 mm × 21 mm as compared to a size of 20 mm × 15 mm on CT.

Common complications of MM are recurrent bacterial infections and renal insufficiency, owing to precipitation of monoclonal light chains in the collecting tubules.^[2,9] Jaw complications include osteolytic lesions causing an increased risk of pathological fractures after oral surgical interventions as well as increased tendency for bleeding.^[3,10]

Conventional dose chemotherapy with melphalan, cyclophosphamide and glucocorticoids, new supportive therapies such as bisphosphonates and erythropoietin, stem cell transplantation and use of bortezomib have proved to be successful in treatment of MM.^[4,10-12] The patient was referred to the higher centers and was put on thalidomide, dexamethasone and zoledronic acid. At 4 months of follow-up patient was found to have no new lesions.

Dental problems in patients of MM should be addressed with the following in the background-tendency of bleeding, poor wound healing, infections, impaired renal function, back pain secondary to involvement of spine, anemia, fatigue, adrenal insufficiency secondary to corticosteroid therapy, osteonecrosis of jaws secondary to bisphosphonates.^[13]

β2 microglobulin, C-reactive protein levels and detection of cytogenetic abnormality like chromosome 13 deletion in cytologic specimens are the prognostic indicators of the disease.^[2] In the present case, a β2

microglobulin level of 3.1 mg/L was suggestive of Stage-I disease with better prognosis.

In conclusion, MM accounting for 1% of all malignant neoplasms, can manifest primarily in the jaws and easily mistaken for a jaw lesion like odontogenic cyst/tumor.^[4] The diagnostician must be aware of the jaw manifestations of MM to prevent potential complications such as bleeding and pathological fracture. Improved diagnostic and therapeutic options along with modern imaging particularly MRI have resulted in improved survival. MRI is superior to CT in depicting the size of the focal lesion.

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