Case Report

Solitary plasmacytoma of the mandible: A rare case report

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ABSTRACT

Plasmacytoma is a monoclonal, neoplastic proliferation of plasma cells that usually arises within bone marrow or soft tissue sites. It can involve either a single bone (solitary) or multiple bones.Solitary plasmacytoma has a predisposition for the red marrow-containing axial skeleton and is most frequently seen in the thoracic vertebrae, followed by the ribs, sternum, clavicle, or scapula. Its presence in the jaws is extremely rare. We present a case of a 54-year-old female with a well-defined radiolucency of the body region of the mandible later diagnosed as solitary plasmacytoma.

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Key words: Mandible, myeloma, plasma cell, solitary plasmacytoma

Introduction

Plasma cell neoplasms are monoclonal neoplastic proliferations of B-lymphoid cells. The course of the disease can be restrained to one medullary area (solitary bone plasmacytoma (SBP)), one extramedullary (soft tissue) area (extramedullary plasmacytoma), or it can involve many areas (multiple myeloma).[1] Solitary plasmacytoma may be an isolated disease or the first manifestation of a subsequent multiple myeloma. Solitary bone plasmacytomas account for about 3-7% of the cases of multiple myeloma.[2,3] Clonal plasma cells involved in plasmacytoma frequently produce a monoclonal immunoglobulin as well as κ or λ light chains. The isolated form of plasmacytoma seems to have a better prognosis, while in cases of subsequent multiple myeloma, the prognosis may be poor. The mean age for solitary plasmacytoma of bone is 55 years, and it shows three times greater male predilection.[4]

Spinal disease is observed in 34-72% cases. Thoracic vertebrae are most commonly involved, followed by lumbar, sacral, and cervical vertebrae. The ileum, humerus, femur, rib, sternum, clavicle, or scapula is involved in 20% of cases. Spinal disease may sometimes be discovered during routine radiography for other conditions, or in case the patient presents with painless swelling of the sternum, rib, or other bones.[4,5] The jaws are rarely involved, with more predisposition toward the mandible. The most common symptom is pain at the site of the skeletal lesion due to bone destruction by the infiltrating plasma cells. Pathologic migration of teeth associated with swelling and paresthesia are observed in the case of larger lesions infiltrating into the neurovascular bundle.[6] In this report, we present a case of a 54-year-old female with a periapical radiolucent lesion in the left body region of the mandible later diagnosed as plasmacytoma.

Access this article online

Quick Response Code:

Website: www.njms.in

DOI: 10.4103/0975-5950.168214

Case Report

A 54-year-old female reported to our unit with the chief complaint of pain and swelling in the left lower back jaw region over the past 2 months. The pain was dull and nonradiating in nature and the swelling had gradually increased over the past 2 months. The patient's medical history was not relevant and she had previously got her tooth of the left lower jaw treated in a private clinic. The general systemic examination was within normal limits. Extraoral examination did not reveal any obvious facial asymmetry or swelling. On palpation, mild tenderness was found to be present over the left body region of the

mandible. Cervical lymph nodes were not palpable and the temporomandibular joint (TMJ) examination was normal.

On intraoral examination, diffuse swelling of about 3 × 2 cm was found involving the gingivobuccal region, involving 35, 36, and 37. The swelling was firm and nonfluctuant, with a slight, deep bluish discoloration of the overlying mucosa[Figure 1]. There was mild tenderness present on palpation. On percussion, 35, 36 and 37 were nontender. Orthop antomogram (OPG) revealed a well-defined 3 × 3-cm periapical radiolucency involving the roots 35, 36, and 37. Of them, 35 appeared to be endodontically treated and with a replaced crown [Figure 2]. Computed tomography (CT) scan showed buccal cortical expansion along with loss of trabeculae and slight perforation of the lingual cortical plate [Figure 3]. On the basis of clinical and radiological findings, a provisional diagnosis was made of unicystic ameloblastoma.

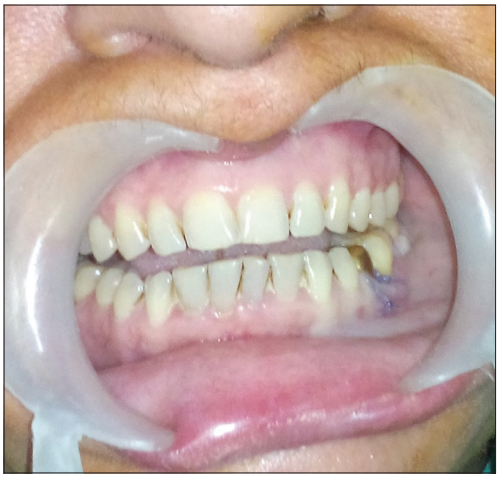

Figure1: Intraoral photograph showing diffuse edema and bluish discoloration of gingivo-alveolar mucosa over the left body region of the mandible

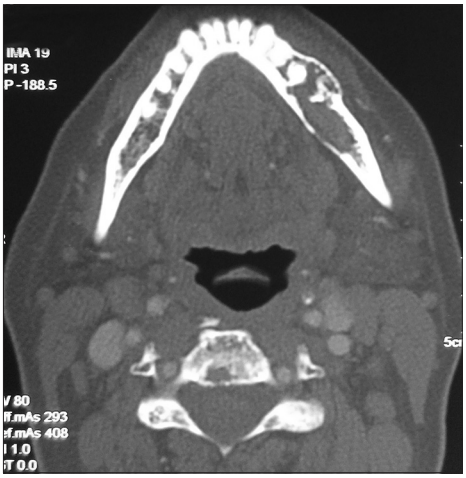

Figure 3: CT scan showing buccal cortical expansion, along with loss of trabeculae and slight perforation of the lingual cortical plate

Treatment was planned for the excisional biopsy of the lesion under local anesthesia (2% lignocaine with adrenaline). The lesion was completely excised, and extraction of 35, 36, and 37 was done [Figure 4]. The tissue was sent for histopathological examination. The histopathological study showed sheets of mature plasma cells along with immature and nucleolated cells permeating the bone, suggestive of plasmacytoma/myeloma, which surprised us [Figure 5]. Immunohistochemical (IHC) markers CD45 or leukocyte common antigen (LCA), epithelial membrane protein (EMA), and CD138 were positive. CD20 was found nonimmunoreactive in plasma cells. Protein electrophoresis of serum showed a characteristic M-spike (monoclonal gammopathy) in the gamma globulin region and hypoalbuminemia. Immunofixation electrophoresis (IFE) of serum identified the M-spike as immunoglobulin G (I_gG) kappa. High-resolution urine protein electrophoresis also revealed the presence of densely staining monoclonal gammopathy (the M-spike) in the gamma globulin region. Notably, x-rays of the skull, chest, lumbosacral spine, and pelvis showed normal findings. All the above investigations were conclusive of solitary plasmacytoma of the mandible. The patient was referred to the radiation oncology unit for adjuvant radiotherapy. Initially she responded

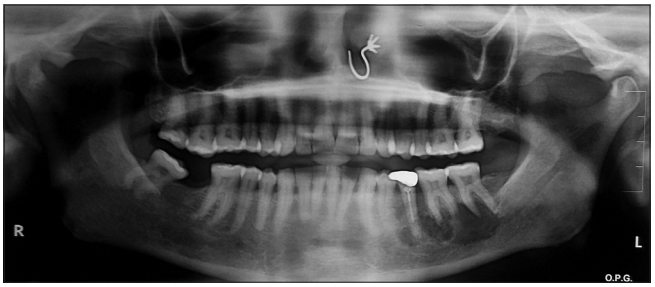

Figure 2: Orthopantomogram showing well-defined 3×3-cm periapical radiolucency involving the roots 35, 36, and 37. Of them, 35 appeared to be endodontically treated and with a replaced crown

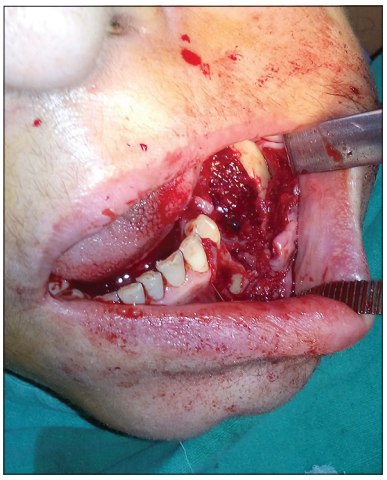

Figure 4: Intraoperative photograph showing the defect after removal of lesion

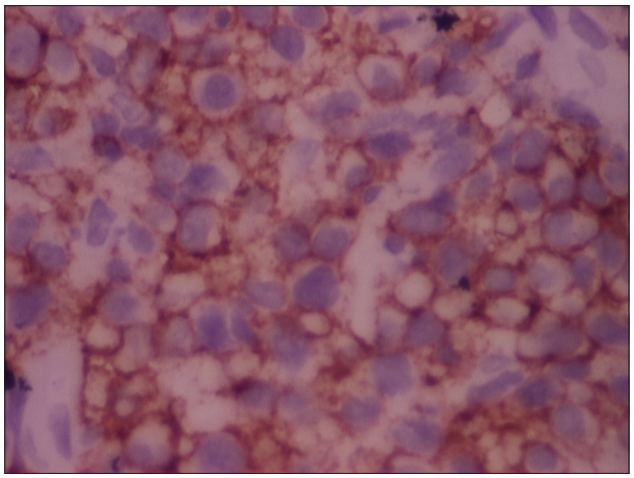

Figure 5: Photomicrograph revealing sheets of mature plasma cells along with immature and nucleolated cells permeating the bone, suggestive of plasmacytoma/myeloma

well, but did not turn up for further follow-ups due to postradiation complications.

Discussion

The jaw is a very rare site of localization of solitary plasmacytoma of the head and neck; symptoms may be underestimated because they are considered nonspecific. Therefore, clinical suspicion of solitary plasmacytoma is difficult, and particular aspects may differ in laboratory and histological findings. Specific symptoms in the case of which to suspect a diagnosis of solitary plasmacytoma localized in a unique bone segment arepain, impairment of bone function and movements, presence of bone swelling with or without local erosion, involvement of local mucosa or tissues, and lack of other systemic symptoms. The clinical presentation of solitary plasmacytoma of the jaw without any pain is very rare, but it has been reported.[7]

The early diagnosis of solitary plasmacytoma must be based on characteristic radiological features, local neurological symptoms, and some systemic manifestations of multiple myeloma that may become evident in solitary plasmacytoma too. Although the reduction of other immunoglobulins is not visible in plasmacytoma, it is possible to find a short peak of gamma globulins in electrophoretic analysis.[7,8] Bone marrow aspirate and/or bone marrow biopsy are normal in solitary plasmacytoma, and are usually diagnostic of multiple myeloma (the presence of more than 15% neoplastic plasma cells). Therefore, only a localized bone biopsy or surgical approach directed toward damaged bone allows the identification of a solitary plasmacytoma. On the other hand, other systemic alterations such as hypocalcemia, hypercalcemia, anemia, leukopenia, thrombocytopenia, and impaired renal function are absent in solitary plasmacytoma, but are usually present in multiple myeloma.[9,10]

The current criteria to make a diagnosis of solitary plasmacytoma are the following: Isolated area of bone destruction due to clonal plasma cells, bone marrow plasma cell infiltration not exceeding 5% of all nucleated cells, absence of further osteolytic bone lesions or other tissue involvement (i.e., no evidence of systemic plasmacytoma); absence of anemia, hypercalcemia, or renal impairment attributable to myeloma; low concentrations of serum or urine monoclonal protein (i.e., myeloma protein); or preserved levels of immunoglobulins.[1,9]

Some clinical conditions of the oral cavity such as poorly differentiated carcinoma and lymphoproliferative disorders may also exhibit similar microscopic featurestothose seen in plasmacytoma. Hence, the diagnosis of solitary bone plasmacytoma (SBP) requires a solitary bone lesion, with both confirmatory histopathological and IHC analyses with the definite support of hematological investigation. IHC markers such as LCA, EMA, and CD138 are essential to show abnormal hematopoietic activity.[11] CD138 is a marker for plasma cells, plasmablasts, and some immunoblasts. Its main reactivity in hematolymphoid neoplasms includes plasma cell neoplasms and some large B-cell lymphomas. CD 45 is specific for both benign and malignant lymphoid (both B-and T-) cells. LCA does not stain megakaryocytes, erythroid cells, normal myeloid cells, or nonhematopoietic cells. EMA is an excellent marker for most normal and neoplastic epithelia but is also expressed by mesotheliomas, meningiomas, and mesenchymal neoplasms.

Treatment methods indicated in solitary plasmacytoma include local surgery (curettage of the lesion), local irradiation, systemic chemotherapy, or a combination of these methods. According to a long-term outcome analysis reported in the literature, a combined radiation and surgical approach gave good outcomes with a low rate of local recurrence.[1,2] The prognosis of solitary plasmacytoma is good due to its benign nature, but 20% of cases may progress to multiple myeloma.[12]

Conclusion

Solitary plasmacytoma of the jaw bone mostly in the mandible is a rare clinical condition. It is uncommon to suspect a unilocular periapical radiolucency of the mandible as plasmacytoma. But if it gets diagnosed on histological and IHC studies, it should be taken into account to rule out its disseminated form, i.e., multiple myeloma.

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How to cite this article: Sharma NK, Singh AK, Pandey A, Verma V. Solitary plasmacytoma of the mandible: A rare case report. Natl J Maxillofac Surg 2015;6:76-9.

Source of Support: Nil. **Conflict of Interest:** None declared.