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Case Report

Solitary plasmacytoma masquerading as a benign lesion

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ABSTRACT

Plasmacytoma refers to a distinct and localized growth of plasma cells that are neoplastic and monoclonal in nature. Solitary plasmacytomas (SP) can be categorized into two groups based on their location: solitary bone plasmacytoma (SBP), which most commonly occurs in the vertebrae and secondarily in long bones, and extramedullary plasmacytoma (EMP), encompassing SPs found outside the bone. Long bones are frequently affected in solitary plasmacytomas. SBP specifically in the jaw is a rare condition, leading to challenges in diagnosis and often resulting in misdiagnosis. SBP carries a higher risk of progression to myeloma, and the primary treatment option is radiotherapy (RT). By employing RT alone, approximately 30% of SBP patients and 65% of EMP patients can achieve long-term disease-free survival (DFS). This article presents a case of a 40-year-old female patient diagnosed with solitary plasmacytoma of the mandible, with detailed documentation of clinical, radiographic, and histological features.

1. Introduction

Plasma cell dyscrasias encompass a range of conditions characterized by the abnormal growth of a single clone of plasma cells, leading to the production of a monoclonal immunoglobulin.[1] One such condition is solitary plasmacytoma (SP), a plasma cell malignancy that falls between monoclonal gammopathy of undetermined significance (MGUS) and multiple myeloma (MM) on the spectrum of plasma cell disorders.[2] These cases constitute less than 5% of all plasma cell dyscrasias and can be further classified as solitary bone plasmacytoma (SBP) or extramedullary plasmacytoma (EMP) based on whether the involvement occurs in bone or soft tissue. SPB is typically limited to a single bone, accounting for 3%–7% of all plasma cell neoplasms.[3] In the context of head and neck involvement, solitary plasmacytoma of the bone is exceptionally rare and is commonly found in the sinonasal tract. However, if it affects the jaw, it tends to favor the mandible over the maxilla.[4] Reported cases in the literature primarily involve bone marrow-rich areas of the mandible, such as the body, angle, ramus, and retromolar trigone. The diagnosis relies on evidence of plasma cell proliferation through biopsy and the absence of involvement in other bones.[5]

2. Case Presentation

A -40-year-old female patient presented with swelling in the lower front tooth region for three weeks. She had a history of blunt hit three weeks back. She experienced pain, which was dull and non-radiating. Swelling had increased to attain present size with time. She had taken antibiotics and analgesics after consulting with dentist but swelling did not subside. No relevant medical, family, personal history was reported. On extra oral examination there was no obvious facial asymmetry or swelling present. Lymph nodes were not palpable. On intraoral examination

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a diffuse swelling of size 10 x3 cm was present on the anterior mandibular region causing obliteration of vestibule extending mesiodistally from the mesial aspect of 35 to the distal aspect of 46 crossing the midline. Superioinferiorly swelling was extending from alveolar mucosa to the depth of the vestibule Overlying mucosa was appeared normal. On palpation there was mild tenderness, swelling was fluctuant and compressible.

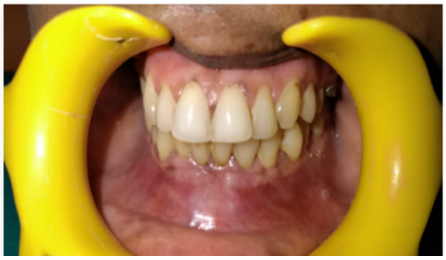

Fig. 1: Intraoral examination revealing A diffuse swelling of size 10 x3 cm present on the anterior mandible causing obliteration of vestibule

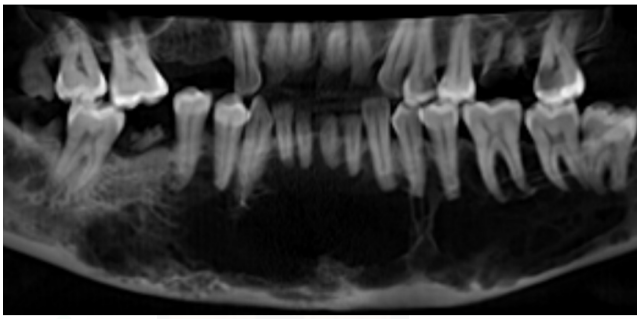

Fig. 2: Orthopantomography revealed solitary well circumscribed multilocular expansile osteolytic lesion present on mandibular anterior alveolus

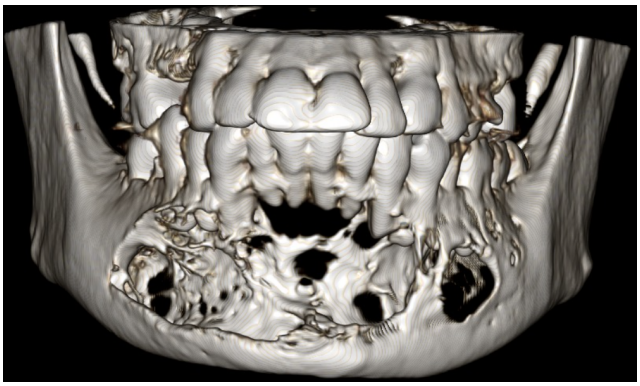

Fig. 3: CBCT revealed expansile multilocular radiolucency extending distal aspect of 36 to distal to 47(95mm) approximately. With faint septae and knife edged root resorption

Based on the provided clinical and radiological findings, a tentative diagnosis of a benign odontogenic tumor was established. However, differential diagnoses including central giant cell lesions, ameloblastoma, and odontogenic myxoma were also taken into account. To gather more information, an incisional biopsy of the bony lesion was carried out under local anesthesia, and the procedure was successfully performed with minimal bleeding.

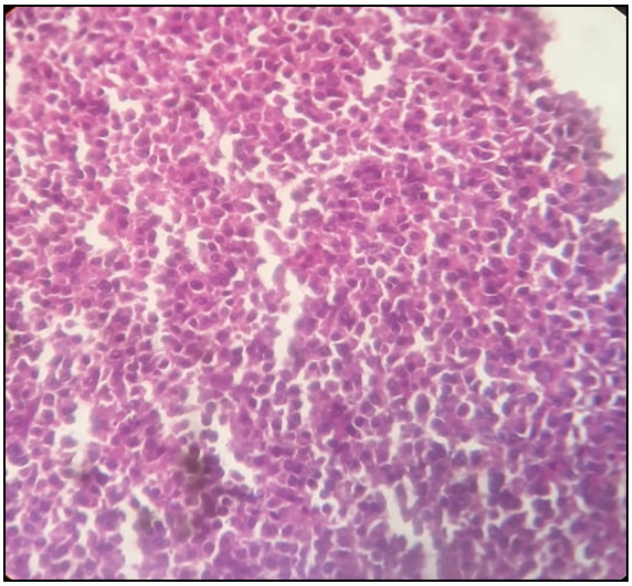

Fig. 4: H&E X 40 showed sheets of plasmacytoid cells and few plasmoblasts

Upon microscopic examination of the H&E stained section, it was observed that the sample contained clusters of closely packed round or ovoid plasmacytoid cells with nuclei positioned eccentrically. A few plasmoblasts were also present. A pink eosinophilic matrix was distributed among the plasmacytoid cells. Based on these findings, a histopathological diagnosis of plasmacytoid non-Hodgkin's lymphoma was made. Differential diagnoses of plasmacytoma and large cell lymphoma of immunoblastic type were considered. To confirm the diagnosis, further laboratory investigations, including routine blood tests, were conducted. In the Complete Blood Count (CBC) test, the white blood cell count (WBC) was 9300 cells/mL, the red blood cell count (RBC) was 4.68 * 10^6 cells/mL, the hemoglobin (Hb) level was 12.2g/dL, and the hematocrit (HCT) was 30.0%. The Mean Corpuscular Volume (MCV) was 74.1 fL, and the mean corpuscular hemoglobin count (MCHC) was 32.8. The MCH was 29.8, suggesting microcytic hypochromic anemia with an increase in rouleaux formation. The platelet count was within the normal range at 330,000/mL. Serum immunoelectrophoresis revealed a mild decrease in serum albumin and a marked elevation in Beta 2 globulin. Monoclonal peaks were detected in the IgA gamma and

kappa regions. The erythrocyte sedimentation rate (ESR) was 120 mm/hr. The albumin-globulin ratio was 0.85.

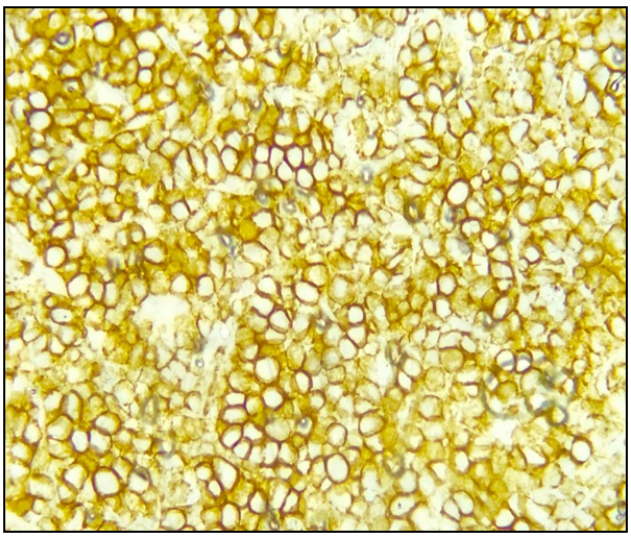

Fig. 5: Immunohistochemistry showing positive for CD138

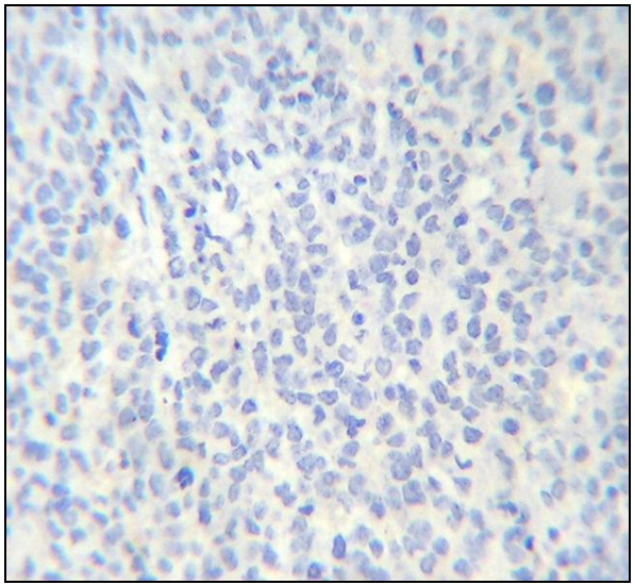

Fig. 6: Immunohistochemistry showing negative for CD 20

To eliminate the possibility of multiple myeloma, a comprehensive systemic evaluation was conducted. Radiographic assessments, including posteroanterior and lateral skull views, were performed, revealing no additional osteolytic lesions. Immunohistochemical analysis demonstrated positivity for CD138 and negativity for CD20. In order to further investigate the potential presence of multiple myeloma, the patient was referred to a hematologist-oncologist for additional tests. A bone marrow aspiration and trephine biopsy were performed, revealing a hypercellular marrow with 6% plasma cells. The patient's serum calcium level was within the normal range, and urinary M protein was not detected. Taking into consideration all the clinical, radiographic, and histopathological findings, the final diagnosis was determined as solitary plasmacytoma of the mandible. The patient was subsequently referred to a specialized cancer center; however, she discontinued treatment after a few weeks and did not return for follow-up visits.

3. Discussion

Solitary plasmacytoma of bone (SPB), as defined by the IMWG, is characterized by the presence of a single lytic lesion caused by the infiltration of monoclonal plasma cells, with or without extension into surrounding soft tissues.[6,7] It is a rare form of plasma cell neoplasm, comprising approximately 5% to 10% of all plasma cell neoplasms according to available literature.[5] SPB is considered an immunoproliferative monoclonal disease that seldom affects the jaw bones, typically presenting as a solitary osteolytic lesion.[5]

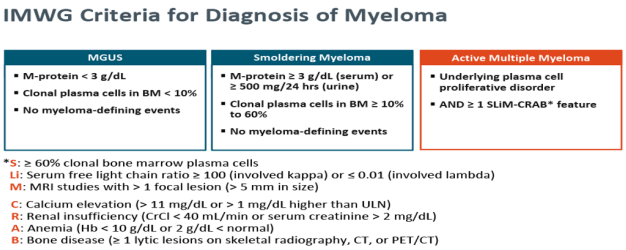

Fig. 7: IMWG criteria for diagnosis of myeloma

The exact cause of solitary plasmacytoma of bone (SPB) remains uncertain, but several hypotheses have been proposed, implicating factors such as radiation exposure, chemical exposure, viral infections, and genetic factors.[1] Cytogenetic studies have shown chromosomal abnormalities including loss of chromosomes 13, 1p, 14q, and gain of chromosomes 19p, 9q, and 1q. Additionally, interleukin 6 is considered a principal growth factor in the pathogenesis of SPB. The incidence rate of SPB is approximately 30% higher in the black race compared to the white race.[8] The median age of patients with either solitary bone plasmacytoma (SBP) or extramedullary plasmacytoma (EMP) is 55 years, and the male-to-female ratio is 2:1. The incidence of SPB increases significantly with advancing age, although it is less prominent in older ages compared to multiple myeloma (MM).[8] SPB primarily affects the axial skeleton, particularly the vertebrae and skull, while involvement of the bones in the extremities is rare. EMP commonly involves the head and neck region, with the nasal cavity and nasopharynx being the most frequent sites.[9] Studies have reported that in most cases, lesions

were observed in the maxilla rather than the mandible.[10] However, there are also reports of anterior mandibular lesions. In this particular case, the lesion was observed in a woman and located in the anterior region of the mandible.[10]

Solitary plasmacytoma of the jaw can manifest with various oral symptoms, including localized pain in the jaws and teeth, paraesthesia (abnormal sensation), swelling, soft tissue masses, mobility and migration of teeth, hemorrhage, and pathological fracture. Fatigue and fever are the most commonly reported systemic symptoms.[4] In the present case, the patient exhibited pain and swelling as the primary symptoms, without any other accompanying manifestations.

On panoramic radiography, the characteristic appearance of solitary plasmacytoma of the jaw is that of an osteolytic image, which can be either single- or multi-locular, without any signs of bone reaction. Computed tomography (CT) scan is essential for a more accurate radiological assessment, providing detailed information about the extension of the lesion into the cortical and soft tissues.[1] According to the 2017 guidelines of the International Myeloma Working Group (IMWG), CT scan is necessary for a definitive diagnosis of solitary plasmacytoma of bone (SPB) on imaging and to evaluate the extent of bone involvement.[6] Lae et al. described three different radiographic patterns observed in SPB, including soap bubble-like multilocular lesions, unilocular radiolucency with a cystic appearance, and ill-defined destructive bone resorption.[5] Our case revealed expansile multilocular radiolucency with faint septae and knife edged root resorption.

Histologically, these tumors consist of well-differentiated plasma cells characterized by small eccentric nuclei with coarse chromatin that appears condensed at the periphery. In some cases, the tumors may also contain immature plasma cells, which exhibit finely dispersed nuclear chromatin within eccentric nuclei, prominent nucleoli, and abundant cytoplasm.[11] Sukpaninchnant et al. described the dysplastic behavior of plasmacytoma, defining minimal dysplasia as the presence of mostly mature plasma cells with less than 10% of cells being plasmoblasts (cells with nucleoli), while marked dysplasia is defined by plasmoblasts accounting for more than 50% of tumor cells.[12] In the present case, the histopathological examination indicated minimal dysplasia, as only a few plasmoblasts were observed.

The current diagnostic criteria for solitary plasmacytoma require both confirmatory histopathological and immunohistochemical (IHC) analysis, along with additional systemic investigations. The criteria include:

Isolated area of bone destruction caused by clonal plasma cells.[1]

Bone marrow plasma cell infiltration should not exceed 5% of all nucleated cells.[1]

Absence of other osteolytic bone lesions or systemic plasmacytoma.[1]

Absence of anemia, hypercalcemia, or renal impairment that can be attributed to myeloma.[1]

Low concentrations of serum or urine monoclonal protein or preserved levels of immunoglobulins.[1]

These criteria help differentiate solitary plasmacytoma from multiple myeloma and other systemic plasma cell disorders.

4. Conclusion

A comprehensive evaluation of signs and symptoms related to systemic disease is crucial in cases of solitary bone plasmacytoma (SBP). It is important to recognize that SBP has a significant risk of progression into multiple myeloma (MM). Therefore, a thorough assessment is necessary to determine the appropriate sequencing of investigations, as this can greatly impact the treatment approach and prognosis for the patient. Identifying any indications of systemic involvement is essential for making informed decisions and providing optimal care.

5. Source of Funding

None.

6. Conflict of Interest

None.

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