



Mandibular plasmocytoma with sun-ray periosteal reaction: A unique presentation

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ABSTRACT

INTRODUCTION: Solitary plasmocytoma is a rare plasmacytic cell tumor, which occurs in the head and neck region and rarely involves the mandible.

PRESENTATION OF CASE: We present a unique radiographic presentation of solitary bone plasmocytoma (SBP) occurring in the Jaw. A 63-year-old male presented with the left mandibular swelling and on the conventional radiograph we noticed a lytic lesion with a sunray periosteal reaction. Clinical diagnosis was osteosarcoma but histopathology revealed sheets of plasma cells with cartwheel appearance and expansile bony trabecula suggestive for solitary bone plasmocytoma. 5 years after complementary treatment by local radiotherapy he developed malaise, weakness and generalized bone pain and bone marrow aspiration revealed more than 90% plasma cell in the marrow and diagnosis of Multiple Myeloma was confirmed.

DISCUSSION: SBP is radiographically seen as a well-defined radiolucent expansile lytic lesion with cortical thinning and no periosteal reaction. The imaging appearance of periosteal reaction is determined by the intensity, aggressiveness, and duration of the underlying pathology. Osteosarcoma, Metastasis (especially from sigmoid colon and rectum), Ewing's sarcoma, Haemangioma, meningioma and Tuberculosis are the main differential diagnosis of Sunburst periosteal reaction.

CONCLUSION: Sunray periosteal reaction should be included in the differential diagnosis of lytic bone lesion in the mandible.

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1. Introduction

A Neoplastic proliferation of B cell lymphocyte is called plasma cell neoplasms. They are divided into three types: disseminated, also called multiple myeloma (MM), localized, also called solitary plasmocytoma of bone (SPB) and extramedullary plasmocytoma (EMP).¹

Solitary plasmocytoma is a rare plasmacytic cell tumor, which occurs in the head and neck region.² It rarely involves the mandible.³ Based on radiologic finding, SBP is a well-defined expansile lytic lesion with thinning of overlying cortex and without sclerosis.

According to our best knowledge, there have been no reports on the periosteal reaction in SBP. In this case report we present a patient with solitary plasmocytoma of mandible with sunray periosteal reaction resembling osteosarcoma "A unique radiologic manifestation".

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Osteosarcoma, Metastasis (especially from sigmoid colon and rectum), Ewing's sarcoma, Haemangioma and rarely fibrous dysplasia are the main differential diagnosis of Sunburst periosteal reaction.⁴

2. Case presentation

A 63-year-old male referred to our hospital due to slowly progressive painful swelling of left mandible of 6-month duration. The symptoms were exacerbated 2 months before admission. Physical examination revealed a left mandibular mass approximately 6 cm in diameter with an osseous consistency covered with a normal skin, without erythema and tenderness. Tongue, lips, buccal, alveolar mucosa and remnant teeth were normal in appearance. His past medical history was unremarkable. He had no previous surgery. His first panoramic radiography of the jaw revealed a lytic lesion in the left mandibular body, 5 mm × 4 mm in dimension, ill defined, without marginal sclerosis, causing sunray appearing periosteal reaction (Fig. 1).

The mandibular and skull radiograph showed no evidence of additional radiolucent lesions. Bone marrow aspiration and serum electrophoresis were normal. Laboratory data such as Hemoglobin, Erythrocyte Sedimentation Rate, Blood Urea Nitrogen, Creatinine,

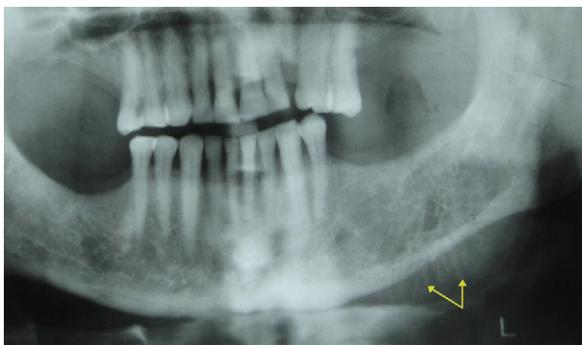


Fig. 1. Panoramic X-ray revealed expansile ill-defined lytic lesion in the left hemimandible with sunray periosteal reaction and cortical disruption (arrow).

Calcium, Phosphorous and Alkaline Phosphatase were within normal limits.

A maxillofacial surgeon performed radical excision of left mandibular body with mandibular reconstruction. The patient was discharged 10 days after surgery without surgical complication and started a liquid diet after 12 days of surgery.

Histopathology revealed sheets of plasma cells with cartwheel appearance and expansile bony trabeculae (Fig. 2A and B) suggestive of solitary plasmacytoma.

Complementary treatment was performed by local radiotherapy with a dose of 45 Gy. The patient was asymptomatic for 5 years after treatment. After the treatment was discontinued he



Fig. 3. Lateral skull X-ray revealed typical (punched out) well defined varying size lytic lesion.

developed malaise, weakness and generalized bone pain and further investigation revealed patients anemia (Hemoglobin: 9 g/dl) and thrombocytopenia (Platelet: 67,000). Radiographic Bone survey revealed typical paunch out lesion in the Skull X-ray (Fig. 3).

Bone marrow aspiration revealed more than 90% plasma cell in the marrow and diagnosis of Multiple Myeloma was confirmed. The patient underwent systemic chemotherapy with malphalan and prednisolone for another 2 years and the disease was controlled. Patient is currently in good health and there is no evidence of disease recurrence.

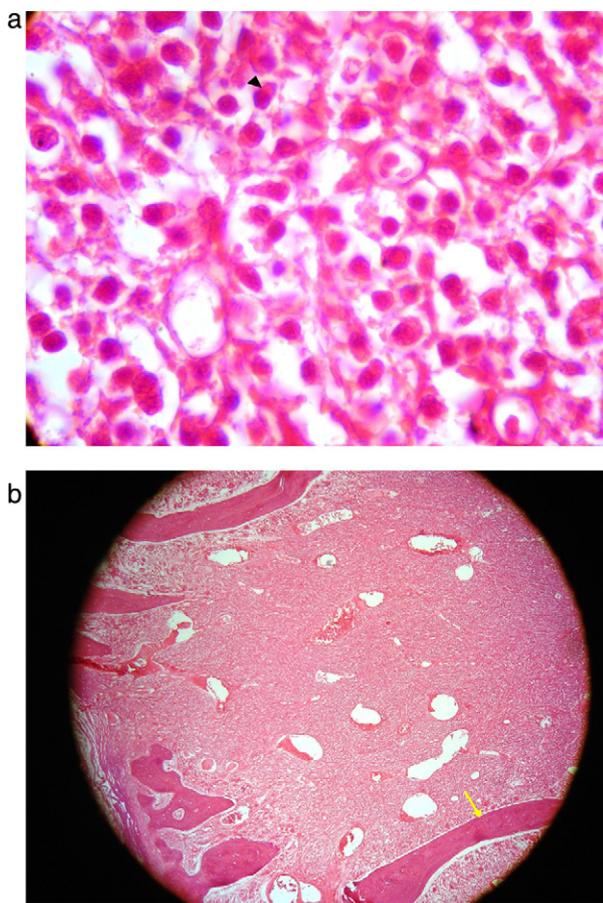


Fig. 2. (A) Hematoxiline and eosin staining shows sheets of plasma cells with cartwheel appearance nucleus (arrowhead) and large nucleus in comparison with cytoplasm. (B) Expansile perpendicular bony trabeculae radiating parallel to each other compatible with sun-ray trabeculae (arrow).

3. Discussion

SBP (3–10% of all plasma cell tumors) is a localized malignant B cell neoplasm. The peak incidence is in the sixth decade and is more prevalent in males (male to female ratio of 2:1).⁴ Skull, Vertebrae and long bones are the most common bony involvement in SBP with maxillofacial involvement in extremely rare reports.^{2,3,5}

SBP is radiographically seen as a well-defined radiolucent expansile lytic lesion with cortical thinning and no periosteal reaction.^{6,7} The differential diagnoses of SBP's are fibrous dysplasia, brown tumor and giant cell tumor. Radiotherapy combined with extensive surgical site excision is the treatment of choice.⁸

Approximately 50% of SBP will transform to MM after several years,¹ which is happened in our patient. Some consider the SBP as an early stage of the disseminated plasma cell neoplasm (MM). Solitary plasmacytoma has a benign course with 10 years survival of approximately 50–80%.⁹

Periosteal new bone formation (“periosteal reaction”) can be due to multiple causes: tumor, infection, trauma, certain drugs, and some rheumatoid disease.¹⁰

According to our knowledge periosteal reaction in SBP has not been reported previously.

The imaging appearance of periosteal reaction is determined by the intensity, aggressiveness, and duration of the underlying pathology. With slow-growing processes, the periosteum has enough time to respond to the disease process and solid, continuous periosteal reaction occurs. In rapidly growing bone pathology, the periosteum cannot produce new bone as fast as the growing lesion. Therefore, a discontinuous rather than a solid pattern of bone formation is seen.¹¹

The periosteal reaction is classified into different subtypes: thin, solid, thick irregular, septated, laminated (onion skin), perpendicular/hair-on-end, sunburst, disorganized, and Codman's triangle. Because dissimilar disease process can provide similar types of periosteal reaction, the major goal of bone radiology is

to recognize the presence of periosteal reaction rather than identifying specific subtypes.

When benign or malignant growth is considered, a discontinuous periosteal new bone formation is usually indicative of a more actively evolving process. These patterns include: Codman's triangle, thin linear opacities perpendicular to the bone (hair-on-end), and speculated linear opacities fanning out from the bone (sunburst).¹²

The rapid growth of the pathologic process prevents confluent maturation of cell elements in the sub-periosteal space in the bone matrix. The matrix deposits along Sharpey's fibers, which supports the periosteum and maintains relationship to the host bone cortex along periosteal vessels.¹³

In the sunburst subtype of periosteal reaction, the spicules of new bone radiate in a divergent pattern instead of being perpendicular (hair-on-end) to the cortex.

Osteosarcoma, Metastasis (especially from sigmoid colon and rectum), Ewing's sarcoma, Haemangioma, meningioma, Tuberculosis, Tropical ulcer and rarely fibrous dysplasia are the differential diagnosis of Sunburst periosteal reaction.⁴

Shozushima et al.¹⁴ reported the first case of multiple myeloma with sunray periosteal reaction in the mandible.

4. Conclusion

In conclusion, although rare, sunray periosteal reaction should be included in the differential diagnosis of lytic bone lesion in the mandible.

Conflict of interest statement

None.

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None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy

of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

Afshin Mohammadi: data collections, data analysis, writing; Behrooz Ilkhanizadeh: design; and Mohammad Ghasemi-rad: writing and editing.

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