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### Cover Page Footnote

SOLITARY BONE PLASMACYTOMA IN MANDIBLE BONE: A CASE REPORT Erwin Danil Yulian, Andre Setiawan Division of Oncology Surgery, Departement of Surgery, Faculty of Medicine Universitas Indonesia

## A Solitary Bone Plasmacytoma In the Mandible: A Case Report

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### Abstract

Solitary bone plasmacytoma is a localized neoplasm in the bone consisting of abnormal plasma cells. We herein report an unusual presentation of solitary bone plasmacytoma in the mandible. A 57-year-old woman with the lump at the right jaw, which was increased gradually for 13 years. The diagnosis build based on clinical, radiological, and histopathological findings. She underwent radiotherapy before surgery using a 50 Gy of irradiation for 25 times results in a partial response. A hemimandibulectomy proceeded, followed by immediate reconstruction using a free vascularized fibular flap. Complete resection may eliminate monoclonal proteins in serum and reduce disease progression into multiple myeloma. The patient remains to follow up regular visits to the oncology surgery clinic in RSCM, with no recurrence of disease in six months postoperative.

**Keywords:** plasmacytoma, mandible, irradiation, mandibular osteotomy

### Introduction

Plasmacytoma is an abnormal growth of  $\beta$  cells of lymphoid plasma cells. It found in two forms: solitary plasmacytoma and multiple myeloma, according to The International Myeloma Working Group (IMWG) in 2003. The first mention form of local abnormality from plasma cell neoplasms, whereas the second one referred to a systemic form. Solitary plasmacytoma divided into two, namely solitary bone plasmacytoma and solitary extramedullary plasmacytoma. Solitary bone plasmacytoma is located in the bone, while the solitary extramedullary plasmacytoma located in the soft tissue. Solitary bone plasmacytoma is an infrequent disorder, representing 5-10% of all plasma cell abnormalities.<sup>1,4,11,12,30</sup> Solitary bone plasmacytoma in the mandible bone was the rarest case; this case is known to be the most common in vertebral bone by 42-61%, pelvic bone by 15%, rib by 12%, lower extremity bone by 12%, upper extremity bone by 10%.<sup>4</sup> Solitary bone plasmacytoma of the mandible reported only in 4.4% of all cases of solitary bone plasmacytoma. If not appropriately managed, solitary bone plasmacytoma may develop into multiple myeloma, which has a poor prognosis with a survival rate of less than two years.<sup>2</sup> A combined treatment modality, which includes radiotherapy and resection, is known to be effective controlling local tumors and suppressing multiple myeloma's progression rate. We herein report a case of solitary bone plasmacytoma in mandible which got combined treatment of resection and radiotherapy.<sup>3,4</sup>

### Case Report

A 57-year-old woman lives in Flores; East Nusa Tenggara presented with swelling at lower right mandible, which progressed slowly for 13

years, from the marble-sized lump to tennis-ball-sized swelling. No pain nor paresthesia, but difficulty in chewing and mouth opening caused by the swelling. No other complaints on visual components, hearing, and smell, nor weight reduction.



Figure 1. Clinical presentation of solitary bone plasmacytoma in the mandible

The extraoral examination showed the lump of 9 cm x 7 cm x 8 cm sized, uneven surface, no different color around the skin, nor ulcer around the lump, well-defined, hard in the consistency, immobilized, no pain on palpation, similar temperature to the surrounding area. No lymph node enlargement identified. The intraoral examination showing partial tumor lump inside the oral cavity with intact mucosa was intact. No ulcer found.

An incisional biopsy carried out with the intraoral approach. The specimen includes the bony mandible and soft tissue as well – the pathology indicated plasmacytoma. The specimen showing abnormal plasma cell proliferation with round-oval shape, pleomorphic, and grainy-chromatin nucleus scattered diffusely distributed—the surrounding soft tissue-restricted tumor cell growth.

Laboratory findings were in normal range with Hemoglobin content of 11.2 g/dL, serum ureum of 16 mg/dL, serum creatinin of 0.6 mg/dL, serum calcium of 9.1 mEq/L. Bence Jones protein in 24-hour urinalysis negative. From blood electrophoresis test there was increased beta-2 of globulin fraction with monoclonal image.

Three dimensions of facial bone CT-scan proceeded preoperatively. The scan is showing a solid mass from the mandible showing a lytic lesion measuring 9.9 cm x 7 cm x 8.8 cm. A destroyed right condyle was identified, while as ramus, angular, and corpus extended into the soft tissue, including the mandible's muscular system. There was no lymph nodes enlargement identified in CT assessment.



Figure 2. The 3D facial CT Scan showing extensive destruction of the hemimandible.

The bone survey was normal; there were no other abnormalities on other bones. Bone marrow puncture showed no abnormal growth of the plasma cell. The Immunohistochemistry (IHC) examination showing positive Lamda, negative Kappa, and positive CD38. The immunoserology tests from blood specimens showing positive gammopathy monoclonal IgG kappa, free light chain kappa of 150 mg/L, free chain lambda of 22.8 mg/L, and kappa/lambda ratio of 6.58.

The intervention to this case consists of the radiotherapy, tumor resection post-radiotherapy right subtotal mandibulectomy. The treatment initiated with radiotherapy with 50 Gy doses of radiation for 25 times; irradiation was given was 2 Gy each. To this irradiation, a partial response is shown. Right subtotal mandibulectomy proceeded following irradiation. The mandibular defect reconstructed using a free vascularized fibular flap.

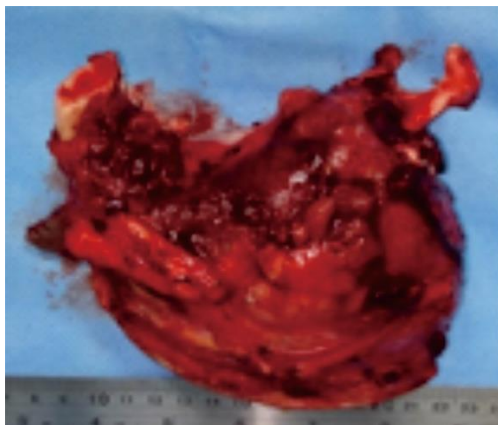


Figure 3. A subtotal hemimandibulectomy proceeded in removing the tumor completely.

After three months follow up, the patient remains regular visits to the oncology surgery clinic in RSCM. Blood specimens showed no progressivity to multiple myeloma. The bone survey showed no abnormalities either.



Figure 4. Clinical presentation in two months after hemimandibulectomy and reconstruction using free vascularized fibular flap.



Figure 5. Intraoral presentation in two months after hemimandibulectomy and reconstruction using free vascularized fibular flap.

## Discussion

Solitary bone plasmacytoma of the mandible bone reported in 4.4 % of the overall cases.<sup>1,4,11,12,32</sup> A multimodality of test required for the diagnosis. A complete blood count, renal function, blood calcium levels. The specific test required is the immunoelectrophoresis-serum test, quantitative examination of immunoglobulin, bone marrow puncture, urine Bence Jones protein, and serum lactate dehydrogenase, and  $\beta 2$  immunoglobulin. The other essential investigations, including bone scans and such as CT Scans, MRI, or PET Scans.<sup>5,16</sup> Diagnostic criteria of IMWG in 2018 are: 1) There are one lytic bone lesions associated with monoclonal plasma cell infiltration in the radiology with or without extension into the soft tissue, and plasma cell neoplasms confirmed results of the histopathologic examination. 2) Clinical and radiological found only a single lesion of bone examination survey. 3) There is no description of plasma cell neoplasms in the bone marrow examination. There is a picture of plasma cell neoplasm in bone marrow examination with a percentage of less than 10%. 4) There is no damage to other organs. 5) There are no systemic symptoms such as anemia, hypercalcemia, and impaired renal function. 6) There is no monoclonal

protein in the urine or serum or monoclonal protein found in urine and serum with a quantity of less than 2.0 g/L.

The mandible is an unusual site for this tumor to grow. Should it grows in the mandible, the resection for local tumor control will be more straightforward. Hemimandibulectomy is done with consideration to achieve complete resection of the tumor according to the oncology's rules, and it shows good aesthetics and function for postsurgery. Complete resection of plasmacytoma tumor may eliminate monoclonal proteins in serum and can reduce the progression of the disease into multiple myeloma.<sup>9</sup>

Solitary bone plasmacytoma is a radiosensitive tumor. The recommended dose of radiotherapy for the management of solitary bone plasmacytoma is 40 Gy, which is given 20 times if the tumor size is less than 5 cm, and a minimum of 50 Gy, which is given 25 times if the tumor size is more than 5 cm. The radiation margin is at least 2 cm from the tumor boundary. Given this therapy, it is known that the local recurrence rate is less than 10%, and the rate of progression to multiple myeloma in 10 years decreases to 30-50%.<sup>6,9,18,19,20</sup>

The tumor response to radiotherapy may found in vary. In common, giving a radiation response of 93% with 62% complete response and 31% partial response.<sup>14</sup> It found that various factors influencing the success rate of radiotherapy and increasing the rate of progression of the disease to multiple myeloma, namely: 1) Patients' age over 60 years 2) Tumor size more than 5 cm. 3) Monoclonal proteins in serum and urine after radiation. 4)  $\beta_2$  microglobulin of more than 3.5 mg/L in serum after radiation. 5) Location of bone lesions in the vertebral bones. 6) Plasma cell growth in pre-radiation bone marrow examination.<sup>3,4,5,6,9,14,21</sup>

Study of Bataline and Sony enrolling 114 solitary bone plasmacytoma cases, which stated that the patients who underwent surgery and radiotherapy had the lowest incidence of multiple myeloma in the future compared with patients who received radiotherapy or surgery alone. A study conducted by the Cancer Center of Sun Yat-Sen University of Korea also supported this statement.<sup>7,8,9</sup> The comparison of local controls of each group was: 5-year local control of patients who received radiotherapy alone was 75%, 5-year local control of patients who received surgery only was 71.4%. In contrast, in solitary bone plasmacytoma patients receiving radiotherapy combined with surgery, the highest five-year local control was 84.6%.<sup>17</sup>

Irradiation, which is initially the primary management, can be a function of postoperative adjuvant therapy to produce higher local and control numbers, and reduce the progression of the disease to multiple myeloma in the future. Therefore, if a case of solitary bone plasmacytoma is found in the mandible, surgery should be carried out before radiation. The tumor in the mandible can be excised completely and may eliminate monoclonal proteins in serum, reduce the progression of the disease into multiple myeloma.

## Disclosure

Authors disclose no conflict of interests

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