

#### Available online at www.sciencedirect.com

## **ScienceDirect**





## **Case Report**

# Solitary plasmacytoma of mandible: A case report \*,\*\*

Zainab El zouiti, MD<sup>a,\*</sup>, Chaimae Amezian, MD<sup>a</sup>, Fahd Elayoubi, MD<sup>b</sup>, Adil Eabdenbi Tsen, MD<sup>a</sup>

- <sup>a</sup> Department of Oral and Maxillofacial Surgery, Mohammed VI University Hospital, Faculty of Medicine and Pharmacy, Oujda, Morocco
- <sup>b</sup> Department of ENT and Cervicofacial Surgery, Mohammed VI University Hospital, Faculty of Medicine and Pharmacy, Oujda, Morocco

#### ARTICLE INFO

Article history: Received 2 September 2023 Revised 15 September 2023 Accepted 20 September 2023

Key words: Case report Mandible Solitary plasmacytoma

#### ABSTRACT

Solitary plasmacytoma is a rare malignant tumor, presenting less than 5% of all plasma cell proliferation. Bone forms are the most frequent, affecting particularly the axial bone skeleton, mandibular localization is extremely rare. Diagnosis is based on the presence of a localized plasma cell tumor without signs of a disseminated form. We report a case of 65 years old female patient with solitary bone plasmacytoma of mandible, who has undergone surgical treatment without adjuvant therapy, with a good clinical and radiological outcomes at 12 months follow-up.

© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

#### Introduction

Solitary plasmacytoma SP is a rare malignant tumor that belongs to a group of large entity of plasma cell dyscrasia, with no systemic involvement [1].

Solitary plasmacytoma regroups 2 different subtypes: SEP or solitary extramedullar plasmacytoma and SBP solitary bone plasmacytoma, it is important to distinguish that the first one occurs originally from soft tissues and tend to sit preferentially in the neck and head area while SBP is present in axial bone skeleton rarely in the jaw [2].

SBP can mostly occur between the fifth and eighth decade of life [3], with a net masculine preference (Sex ratio 2/1) [4].

Clinical presentation is characterized by the presence of voluminous tumefaction, swelling, pain and difficulty masticating [5]. Radiological assessment can show either radiolucent area or lytic mass of the mandible [6,7], the biopsy with histological study confirms the diagnosis [8].

Systemic investigations are necessary once SBP diagnosis is done including whole body imagery, bone marrow biopsy, hematological examination, urine and plasma electrophoresis to search for systemic dissemination [3].

E-mail address: zainab.elzouiti@gmail.com (Z. El zouiti).

https://doi.org/10.1016/j.radcr.2023.09.061

1930-0433/© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Acknowledgments: We have any financial sources.

<sup>\*\*</sup> Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

<sup>\*</sup> Corresponding author.



Fig. 1 - Clinical presentation of the case.

#### Case report

We report the case of a 65-year-old female patient with no medical history, admitted for a voluminous none painful tumor of the lower right jaw evolving for 2 years, clinical examination showed a large lower cheek tumefaction without skin inflammatory signs measuring approximately 7 cm in diameter, at palpation, it was firm, sensitive, depending on the mandibular bone from the body all the way to ramus area. The cervical lymph nodes examination was normal (Fig. 1).

Intraoral examination revealed vestibular expression of the tumoral lesion, with partial toothless of the region in question.

A CT scan was ordered showing a lytic process evolving in the right side of the mandibular body to the ramus area all the way up to Temporo mandibular joint TMJ (Figs. 2-4).

We performed an hemi mandibulectomy with total TMJ removal by extra oral approach (Fig. 5) followed by reconstruction with maxi plate and condylar prosthesis (Fig. 6) the post operative period was uneventful and the patient was discharged at the 4th post operative day with regular follow up.

The histological study returns in favor of a plasmacytoma with free margins. an extension assessment with blood investigations were without any abnormalities including no hypercalcemia, no urinary Bence Jones proteins within the electrophoresis. in order to find other body skeletal lesions a Body CT scan was conducted that comes back without abnormalities.

The final diagnosis of solitary plasmacytoma was given. Our patient did not receive any adjuvant treatment. A 12 months follow up results were good with no signs of recurrence.



Fig. 2 – 3D reconstruction CT scan images showing the destruction of the mandibular bone by the tumor.

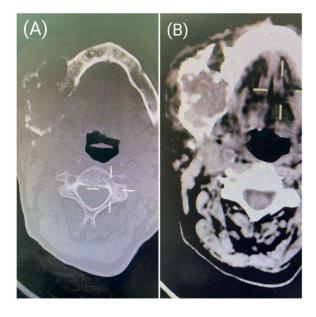


Fig. 3 – Axial CT scan view showing an osteolytic lesion of the mandible with cortical bone destruction (A) bone window (B) soft tissue window.

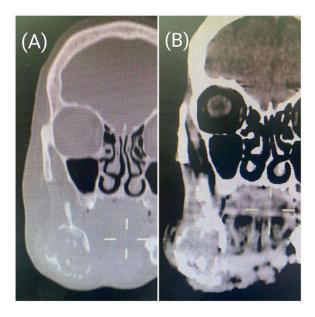


Fig. 4 – Coronal CT scan view (A) bone window (B) soft tissue window.



Fig. 5 - Peroperative aspect of surgical approach.



Fig. 6 - Postoperative panoramic X-ray.

#### Discussion

Solitary plasmacytoma represent less than 5% of plasma cell dyscrasias [1], the most common locations of SBP are vertebrae, long bones, rarely jaws, mandibular location is extremely rare [2].

SBP occurs most frequently in patients between the ages of 50 and 80, rarely before 40 [3], it is more prevalent among men then women with a sex ratio of 2/1 [4].

The etiology of SBP remains uncertain, multiple hypotheses were suggested such us chemical exposure, radiation, viruses, and genetic factors implicated in SBP pathogenesis [8].

Our patient had an increase in the tumor volume as the main symptom, our results align with those found in literature that also identify this sign as the chief clinical sign. Swelling, sensory disturbances can also be described, less commonly pathological fractures [5].

To conclude to SBP diagnosis there must be no other bone/organ involvement including: single area bone destruction, bone marrow infiltration being less than 5% of all nucleated cells. Blood test showing no anemia, no hypercalcemia, low concentration of serum/urine monoclonal proteins nor renal lesion attributed to multiple myeloma, in addition to an histological and immunohistochemical proof [3].

Radiological assessment is not specific, in most studies, the solitary plasmacytoma appears as a diffuse, multilocular radiolucent lesion. CT scan can help precise cortical degree of destruction [6,7].

There over MRI can describe better soft tissues and bone marrow involvement [1].

Other lesions could mimic SPB aspect such as cystic and pseudocystic tumors, in addition to malignant tumors [6,7]. In our case, we almost confused the diagnosis with ameloblastoma based on radiological findings.

The biopsy of the lesion and its histopathological study confirms the diagnosis, dystrophic plasmocyt cells can be associated with nucleus abnormalities with a total normal cytoplasm.

Dystrophy is the key element for histological diagnosis, clonal nature of plasmocyt cells is determined by histochemical study [8].

Therapeutic approach includes several methods: surgical resection, local irradiation, and systemic chemotherapy.

However, radiotherapy remains the gold standard given the radiosensitivity of these tumors, with a satisfactory local control rate which varies between 80% and 90% of cases presented in literature [1].

Surgical resection may be indicated in accessible peripheral locations, and should not be mutilating, given the equivalent efficiency compared to radiotherapy [9].

In mandibular location surgery is required in almost cases, for mainly aesthetic reasons. Adjuvant radiotherapy is still necessary to avoid local recurrence [1].

In our case, adjuvant radiotherapy was not indicated, since the excision was within free margins.

According to latest recommendation, the role of adjuvant chemotherapy remains controversial, it may be indicated in cases with local recurrence, or in patients with high risk of treatment failure [9].

Three types of relapse were observed: development of a multiple myeloma (54%), local relapse (11%), appearance of new lesions (multiple plasmacytomas) in the absence of multiple myeloma (2%). The main predictors of recurrence are tumor size and the dose of radiotherapy delivered, with a higher risk for tumors larger than 5 cm and in case of radiotherapy with a dose less than or equal to 35 Gy [10].

The prognosis of solitary bone plasmacytoma remains essentially dominated by the risk of occurrence of multiple myeloma. Age over 52 years, vertebral seat, existence or persistence of a monoclonal peak 1 year after treatment are the main risk factors [10].

#### Conclusion

Solitary bone plasmacytoma is a rare malignant tumor which mainly affects the axial skeleton, mandibular location is extremely rare. Clinical and radiological findings are not specific. The treatment of choice remains radiotherapy alone at a moderate dose ensuring good local control in 90% of cases. Some cases require surgery especially jaw localization. The prognosis depends on its evolution towards multiple myeloma, requiring regular follow-up and better therapeutic management of patients suffering from signs of systemic dissemination.

### Patient consent

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.

#### REFERENCES

- [1] Pham A, Mahindra A. Solitary plasmacytoma: a review of diagnosis and management. Curr Hematol Malig Rep 2019;14(2):63–9. doi:10.1007/s11899-019-00499-8.
- [2] Seoane J, Aguirre-Urizar JM, Esparza-Gomez G, Suarez-Cunqueiro M, Campos-Trapero J, Pomareda M. The spectrum of plasma cell neoplasia in oral pathology. Med Oral 2003;8(4):269–80.
- [3] Pisano JJ, Coupland R, Chen SY, Miller AS. Plasmacytoma of the oral cavity and jaws: a clinicopathologic study of 13 cases, Oral Surg. Oral Med 1997;83(2):265–71. doi:10.1016/s1079-2104(97)90015-9.
- [4] EM Canger, P Çelenk, A Alkan, and O Günhan, "Mandibular involvement of solitary plasmocytoma: a case report," Med Oral Patol Oral Cir Bucal, 12:E7-9.
- [5] "Florencio M, Jose L, Gil-Diez Campano FJ, Jose R, Del Hoyo A. Mandibular lesion as the first evidence of multiple myeloma. J Craniomaxillofac Surg 1989;17:315–17.
- [6] Bellalah Z, Slama A, Ayachi S, Rammeh S, Mrad-Daly K, Khochtali H. Plasmocytome solitaire de la mandibule. A propos de deux cas. Actual Odonto Stomatol. 2010(250):145–51. doi:10.1051/aos/2010205.
- [7] Lesmes D, Laster Z. Plasmacytoma in the temporomandibular joint: a case report. Br J Oral Maxillofac Surg 2008;46:322–4.
- [8] Chittemsetti S, Guttikonda VR, Sravya T, Manchikatla PK. Solitary plasmacytoma of mandible: a rare entity. J Oral Maxillofac Pathol 2019;23(1):136–9. doi:10.4103/jomfp.JOMFP\_175\_18.
- [9] Caers J, Paiva B, Zamagni E, Leleu X, Bladé J, et al. Kristinsson, Diagnosis, treatment, and response assessment in solitary plasmacytoma: updated recommendations from a European Expert Panel. J Hematol Oncol 2018;11(1):10. doi:10.1186/s13045-017-0549-1.
- [10] Hadadi K, Hommadi M, Belemlih M, Zaghba N, Maghous A, Marnouch E, et al. Plasmocytome solitaire osseux: expérience du service de radiothérapie de l'hôpital militaire d'instruction Mohammed-V de Rabat (Maroc). Cancer Radiother 2019;23(8):867–73. doi:10.1016/j.canrad.2019.06.013.