An Oral Plasmacytoid Mass: The Result of a Malcontrolled Multiple Myeloma

Narges Mirjalili, Mohammah-Hassan Akhavan Karbassi, Ali-Akbar Davoudi Department of Oral Medicine, Shahid Sadoughi University of Medical Sciences, Yazd, Iran

Corresponding author: Nargess Mirjalili, Assistant professor in oral medicine

Fazay-e-sabz square, Department of oral medicine, Shahid Sadoughi university of medical sciences, Yazd, Iran

Tel: +3516212222, +989124024930 E-mail: nargess1981@yahoo.com

Co-author one: Mohammad-Hassan Akhavan Karbassi, drkarbassi@gmail.com

Co-author two: Ali-Akbar Davoudi, aadavoudiom@gmail.com

Abstract

Introduction: Multiple myeloma is a neoplasm of monoclonal plasmacells which may cause oral manifestations. We report a 50-year-old man with a mass burned out of a post extraction socket. He had recived an eight month period of chemotherapy and was supposed to be ready for hematopoetic stem cell transplantation. After clinical and histopathological examinations, the mass was diagnosed as a plasmacytoid cell proliferation. As the oral manifestations are an indicator of poorly controlled disease, the patient was refered to change his chemotherapeutic regimen.

Conclusion: It seems that a team work, including oral medicine specialists may be a valuable opportunity in monitoring and promoting the quality of therapy.

Keywords: Multiple myeloma, Oral manifestations, Oral medicine

Introduction

Multiple myeloma represents a neoplastic growth of monoclonal plasmacells.(1) The cancerous cells primarily involve the bone marrow spaces in multiple sites and then may involve exteramedullary tissues.(2) Clinical manifestations of the disease such as pain, pathologic fractures, renal failure and recurrent infections are results of aberrant cell proliferation, neoplastic products and host responses.(2, 3) In head and neck region, radiolucent punched-out or ragged border bone lesions, soft tissue amyloid deposition and intra-oral metastatic tumoral masses may be seen especially as late consequences of disease.(4-7)

Management of patients with multiple myloma is fundamentally directed toward regression of the malignant process and palliation of symptoms.(1,8 9) In past 2 decades, Hematopoetic stem cell transplantation (HSCT) has been vastly used beside conventional or modified chemotherapeutic regimens.(9-13) HSCT is a way to prolong the event free survival of patients and is better to be done after the first successful induction therapy.(3, 14)

Emerging manifestations of multiple myeloma in oral cavity, is a strong indicator of uncontrolled disease progression.(4, 7)

Case presentation

A 50-year-old man with multiple myeloma, attended to Oral medicine center of Yazd medical university for pre-transpant oral health evaluation. His disease was first diagnosed following a spontaneous fracture in his upper exterimity. The patient had recived a chemotherapeutic regimen of melphalan and dexamethasone for eight months and was assumed to be ready for HSCT. In first visit, the patient complained of a mass that appeared 3 weeks ago in his mouth, after a tooth extraction. He mentioned a history of a prolonged deep pain in his right angle of mandible. This situation convinced his oncologist to refer him for dental treatments. Then diagnosing a deep carious tooth in his lower left quadrant, rationalized its extraction.

Intra-oral examinations revealed a non-healing tooth socket and a 1×1.5 cm mass, burned out of the socket. It was rubbery in palpation and fixed to underlying bone. Panoramic radiography merely showed a non-healing post extraction socket. Our differential diagnosis was ordered as below:

- 1. Plasmacytoma/ Multiple myeloma
- 2. Lymphoma
- 3. Central giant cell granuloma

Complete blood cell count, PT/INR and BT tests were requested in order to perform a surgical

diagnostic incisional biopsy. Although the laboratory values were all within normal limits and the patient was supposed to be in remission phase, we encounterd a bleeding situation. Despite the restricted surgical field, the volume of bleeding was enormous and hardly controlled by local hemostatic modalities. The excised tissue was very fragile and suturing of surgical margins was too difficault.

Histopathological study of the obtained specimen indicated diffuse monotonous sheets of variably differentiated malignant plasmacytoid cells which was compatible with plasmacytoma. Further radiographic examinations revealed multiple punched-out lytic lesions of the skull. All the evidences confirmed our first diagnosis and implied the malignant process did not respond to provided chemotherapeutic regimen.

We reffered the patient to his oncologist, recommending to switch the chemotherapy plan. HSCT was carried out after the first remission, about one year past our first visit. The patient is alive by the time of this paper and the oral cavity is diseas free.

Discussion

Multiple myeloma primarily involves the bone in a multi-centeric fashion but is considered a hematologic cancer of monoclonal plasma cell origin.(1) It arises from bone marrow spaces and rarely manifests as soft tissue metastases.(2, 15) Neoplastic cells replace can the normal hematopoetic cell lines, destruct involved bones and waste the body resources by producing aberrant proteins. Accordingly, skeletal pain, pathologic hypercalcemia, anemia, fractures, frequent infections, bleeding tendency and renal impairment are not unexpected in the course of the disease.(1, 16)

In head and neck region, the most common features are osteolytic lesions of skull, cervical spine and posterior mandibular areas which may lead to pain, paresthesia, tooth loosening and fracture.(6, 4, 7, 15) Patients with multiple myeloma, also may manifest plasmabelastic soft tissue masses in oral cavity.(17-19)

In the present case, we encountered a chief complaint of a mass, burned out of a post-extraction socket. It was rubbery in consistency and fixed to underlying tissue. There was also a history of a continous deep pain in that area of posterior mandible. A list of differential diagnosis was arranged based on the clinical findings and medical history. At the lowest rank, one may consider a giant cell lesion as the propable diagnosis. Renal

dysfunction secondary to multiple myeloma, cytokine profile of the disease and abnormal osteoclastic activity, may all disturb the normal bone turnover.(1-3) So it is not impossible if a central giant cell lesion forms. Central giant cell lesions prefer to emerge through less resistant parts of jaw bone into the oral cavity and may bleed heavily during surgical intervantions.(20)



Figure 9- The clinical view: The mass burned out of the dental socket after the extraction.



Figure 10- The panoramic view. The shadow of the mass is evident at the posterior right part of the mandible.

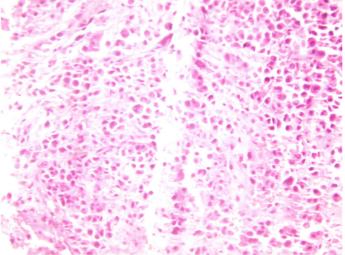


Figure 11- The histopathological view: Large plasmacytoid cells are obvious.

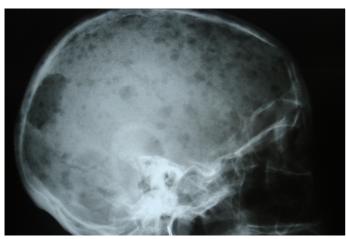


Figure 12- The lateral skull view: Multiple punched out lesions involved the whole skull.

On the other hand, these pathoses usually are firm in palation, frequently involve anterior mandible, expand the involved bone, and are very rare in this context.(20) As the underlying disease would be recognized much more earlier, it seems impossible to progress to such a condition.

The second possible diagnosis could be chemotherapy induced Lymphoma. Cytotoxic drugs usually have low therapeutic indexes. They affect both normal and neoplastic cells. Long term consumers of chemotherapeutic agents are at a greater risk for developing secondary hematologic cancers than normal population.(21)

Involving lower jaw especially in the posterior parts, burning out of an extraction socket, rubbery consistency, fragility and bleeding tendency are all characteristics of extera-nodular lymphomas.(6, 20) This assumption is potentiated when highlighting an eight month use of melphalan in patient's history.

Considering the backround medical status of the patient, it was reasonable to put the extra-medullary plasmacytoid mass in the top rank of our differential diagnosis. The history and the clinical, surgical and histopathological findings were all consistant with the diagnosis of multiple myeloma. Multiple myeloma is a rare malignancy with a wide range of complications.

It is very unexpected to see mouth lesions as the first manifestation of the disease and oral involvemens such as pain, paresthesia of inferior alveolar nerve, tooth loss, osteolytic lesions of jaw, amyloidosis, gingival masses and peripheral plasmacytoid tumors are more prone in progressive or uncontrolled disease.

During the treatment course, high-dose chemotherapeutic drugs may affect both directly and indirectly the mouth, and for a successful HSCT, stabilishing opltimal oral hygine, removeing

infectious sources and providing a healthy oral environment is a critical consideration. So the spectacular role of an oral medicine especialist is underlined in all stages of the disease from diagnosis to monitoring chemotherapeutic regimens efficacy and/or preparing for HSCT.

Conclusion

Considering the available evidences, we suggest that the oral medicine specialists become involved in close monitoring of medical management of patients with multiple myeloma and it would be appreciated if medical care providers use the advantage of our consultations in this regard.

References

- 1. Dennis L Kasper, Tinsley Randolph Harrison. Harrison's Principles of Internal Medicine. 16 ed: McGraw-Hill, Medical Pub: 2005.
- 2. Mahindra A, Hideshima T, Anderson KC. Multiple Myeloma: biology of the disease. Blood Reviews 2010;24(Supplement 1):S5-S11.
- 3. Gertz MA, Ghobrial I, Luc-Harousseau J. Multiple Myeloma: Biology, Standard Therapy, and Transplant Therapy. Biology of Blood and Marrow Transplantation 2009;15(1, Supplement 1):46-52.
- 4. Lambertenghi-Deliliers G, Bruno E, Cortelezzi A, Fumagalli L, Morosini A. Incidence of jaw lesions in 193 patients with multiple myeloma. Oral Surg Oral Med Oral Pathol 1988 65(5):533-7.
- 5. Mozaffari E, Mupparapu M, Otis L. Undiagnosed multiple myeloma causing extensive dental bleeding: Report of a case and review. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology & Endodontics 2002;94(4):448-53.
- 6. Lester William Burket, Martin S. Greenberg, Michaël Glick, Jonathan A. Ship. Burket's oral medicine. 11 ed: PMPH-USA: 2008.
- 7. Seoane J, Aguirre-Urizar J, Esparza-Gómez G, Suárez-Cunqueiro M, Campos-Trapero J, Pomareda M. The spectrum of plasma cell neoplasia in oral pathology. Med Oral 2003 8(4):269-80.
- 8. Stoopler ET, Vogl DT, Stadtmauer EA. Medical management update: Multiple myeloma. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology 2007;103(5):599-609.
- 9. Reece DE. Management of multiple myeloma: The changing landscape. Blood Reviews 2007;21(6):301-14.
- 10. Harousseau J-L, Attal M. The role of stem cell transplantation in multiple myeloma. Blood Reviews 2002;16(4):245-53.

- 11. Hahn T, Wingard JR, Anderson KC, Bensinger WI, Berenson JR, Brozeit G, et al. The Role of Cytotoxic Therapy with Hematopoietic Stem Cell Transplantation in the Therapy of Multiple Myeloma: An Evidence-Based Review. Biology of Blood and Marrow Transplantation 2003; 9:4-37.
- 12. Theresa H, John RW, Kenneth CA, William IB, James RB, Greg B, et al. The role of cytotoxic therapy with hematopoietic stem cell transplantation in the therapy of multiple myeloma: An evidence-based review. Biology of blood and marrow transplantation: journal of the American Society for Blood and Marrow Transplantation 2003;9(1):4-37.
- 13. Pant S, Copelan EA. Hematopoietic Stem Cell Transplantation in Multiple Myeloma. Biology of Blood and Marrow Transplantation 2007;13:877-85.
- 14. Kim JS, Kim K, Cheong J-W, Min YH, Suh C, Kim H, et al. Complete Remission Status before Autologous Stem Cell Transplantation Is an Important Prognostic Factor in Patients with Multiple Myeloma Undergoing Upfront Single Autologous Transplantation. Biology of Blood and Marrow Transplantation 2009;15(4):463-70.
- 15. Segundo AV-L, Falcão MFL, Filho RC-L, Soares MSM, López JL, Küstner EC. Multiple Myeloma with primary manifestation in the mandible: A case report

- Med Oral Patol Oral Cir Bucal 2008;13(4):E232-4.
- 16. Glaspy JA. Hemostatic abnormalities in multiple myeloma and related disorders. Hematol Oncol Clin North Am 1992 6(6):1301-14.
- 17. Castellarin P, Pozzato G, Tirelli G, Lenarda RD, Biasotto M. Oral Lesions and Lymphoproliferative Disorders. J Oncol 2010;2010:1-10.
- 18. Ozdemir R, Kayiran O, Oruc M, Karaaslan O, Koçer U, Ogun D. Plasmacytoma of the hard palate. J Craniofac Surg 2005 16(1):164-9.
- 19. Lee S-H, Huang J-J, Pan W-L, Chan C-P. Gingival mass as the primary manifestation of multiple myeloma: Report of two cases. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology 1996;82(1):75-9.
- 20. Brad W. Neville, Douglas D. Damm, Carl M. Allen, Jerry Bouquot. Oral and maxillofacial pathology. 3 ed: Saunders/Elsevier: 2009.
- 21. Anonymous. Acute leukaemia and other secondary neoplasms in patients treated with conventional chemotherapy for multiple myeloma: a Finnish Leukaemia Group study. Eur J Haematol 2000 65(2):123-7.