Oral Oncology EXTRA (2004) **40** 13–15

ELSEVIER

ORAL ONCOLOGY EXTRA

www.elsevier.com/locate/ooe

CASE REPORT

Burning mouth syndrome as the initial sign of multiple myeloma

V. Vučićević-Boras*, I. Alajbeg, S. Brozovic, M. Mravak-Stipetic

Department of Oral Medicine, School of Dentistry, University of Zagreb, Gunduliceva 5, 10 000 Zagreb, Croatia

Received 20 October 2003; accepted 10 November 2003

Summary A 79-year-old women was referred to our Department with burning symptoms in the whole oral cavity together with clinically healthy appearence of the oral mucosa. Hematological tests as well as bone marrow biopsy revealed diagnosis of plasmocytoma. The aim of our case report was to underline the importance of hematological screening in patients with burning mouth syndrome.

Introduction

Multiple myeloma is a malignant disease that is characterized by multifocal proliferation of atypical plasma cells and, in most patients, by the presence in the serum of monoclonal gamma globulins and/or their subunits, often referred to as M or myeloma proteins. Secondary invasion of skeletal tissue is one of the most important characteristics of multiple myeloma, radiologically evident in about 80% of the cases.[1] It is most commonly seen within the age range 60–70 and men are affected more frequently. Neoplastic proliferation originates primarily in hematopoietic marrow manifesting in anemia, trombocytopenia and neutropenia. In 12–15% of cases of multiple myeloma the first manifestation of the disease appears in the jaw bones and oral cavity.[2] Oral manifestations of multiple myeloma are clinical features such as pain localized in the jaws or teeth, paresthesias, swelling, soft tissue masses, mobility of the teeth, migration of teeth, hemorrhage and pathologic fracture due to osteolytic bone lesions.[3] Malignant myeloma is recognized as a strong suspect for unexplained chronic bone pain in all parts of the skeleton. In the jaws such pain is usually associated with the presence of osteolytic defects, but atypical facial pain can result from sensory neuropathia which occurs in multiple myeloma even when the radiologic examination fails to reveal those bony defects.[4]

Case report

A patient PM, 79 years old was referred to the Department of Oral Medicine in Zagreb, Croatia with symptoms of burning in the whole mouth, especially severe on the anterior 2/3 of the tongue, lasting for two months. The patient was not previously treated. Clinical examination showed healthy appearence of the whole oral mucosa. In the maxilla teeth 21, 22, 23 were present and the

*Corresponding author. Tel.: +385-1-4802-124; fax: +385-1-4802-159.

E-mail address: vvboras@hotmail.com (V. Vučićević-Boras).

1741-9409/$ - see front matter
doi:10.1016/j.ooe.2003.11.003

mandibular alveolar ridge was completely edentolous. The tooth 22 was asymptomatic, but X-ray finding showed diffuse periapical radiolucency around its root. No other bone defects could be seen on X-ray finding. Regional lymph nodes were not palpable. Detailed medical history revealed surgical treatment of gallbladder in 1986, gastric disturbances, and pancreatitis. Glucose tolerance test was normal.The patient was sometimes suffering from vertigo, headaches and she lost 14 kg during past year due to a diarrhoeal episodes. Prescribed medications included hypnotics-benzodiazepin, three times daily (3 mg$\times$3 mg); acetylsalic acid (100 mg once a day), and sometimes alprazolam 1 mg once a day.

Candida culture from oral smears was positive. The smear was taken and placed on Saboraud's agar for next 48 h on 37 °C. Candidal infection was treated with antifungals (2% miconasolum gel four times a day during 10 days) and afterwards severe burning was still persistent.

Routine blood screen tests were performed and showed decreased red blood count being 3.07×10^{12}/l (normal range 3.86—5.08×10^{12}/l), decreased hemoglobin levels 91 g/l (normal range 119—157 g/l), decreased hematocrit 0.272 l/l (normal range 0.356—0.470 l/l), RDW was elevated being 17.9 (normal range 14.6—16.5, individual erythrocyte distribution); platelet count was 120×10^{9}/l (normal range 158—424×10^{9}/l) and neutropenias was present 34.9% (normal range 44—72%). Erythrocyte sedimentation rate was elevated and was 78 mm/h (normal range 5—28 mm/h) according to Westergreen. Due to an abnormal hematological finding she was reffered to hematologist. A bone marrow biopsy specimen revealed abnormal bone remodelling with reduced granulocytopoesis albeit morphologically normal. Proliferation of plasma cells was visible and they made up 75%, which were partially pleomorphic. Both findings were attributed to the diagnosis of plasmacytoma. Bone marrow cytology showed normocellular bone marrow but with erythrocyte, granulocyte and platelet cells merged to the periphery of the bone marrow, and atypical plasma cells and plasmablasts presenting with 57% of all cell types. Renal function was normal, although urinalysis showed high protein content being more than 1.0 g/l while the referent values are less than 0.05 g/l.

X-ray finding of the lungs as well as whole body scan were normal. Electrophoresis of serum proteins showed decreased albumin levels (38.1%, normal 57—69%, normal alpha 1 being 2.7% (2.0—4.0%), decreased alpha 23.1% (5.0—10.0%), increased beta s 51.1% (8.0—12.0%) and decreased gamma being 5.0% (13.0—22.0%). A monoclonal M protein was detected on serum protein electrophoresis and on following immunelectrophoresis was identified as IgG. IgG values were increased (43.5 g/l, normal range 10.0—19.0 g/l) and IgA values were decreased (0.1 g/l, normal range 1.1—3.8 g/l) as well as IgM values (0.1 g/l, normal range 0.7—2.6 g/l). The diagnosis of IgG plasmacytoma was established by hematologist. One year after the multiple myeloma was diagnosed, the patient committed suicide.

Discussion

Oral manifestations may be the first signs or symptoms of multiple myeloma. Epstein et al.[2] reviewed 783 cases from the literature and found oral manifestations in 14% of patients with multiple myeloma. Neuropathy is a dominant feature in only 10% of patients with multiple myeloma, and it commonly preceeds the discovery of the blood dyscrasia.[5] Monoclonal anomalous protein may have the property of antinerve antibody and be able to cause the damage of axons and myelin in nerves and subsequential sensory neuropathy.[6] This case report alert on the possible association of burning mouth syndrome with multiple myeloma, a finding not previously reported in the literature. Previous reports of burning mouth syndrome as a paraneoplastic syndrome were reported in patients with malignant tumors of the pulmonary tract[7] and the acustic nerve neuroma.[8] However, at this point it cannot be discriminated whether burning mouth syndrome is merely a coincidential finding, or it is a result of the multiple myeloma itself. Nevertheless, it is our obligation to underline the importance of hematological screening, such as complete blood count in patients with burning mouth syndrome as a possible marker of blood discrasias particularly malignant diseases of the blood.

References

1. Lambertenghi-Deliliers G, Bruno E, Corteleyyi A, Fumagalli L, Morosini A. Incidence of jaw lesions in 193 patients with multiple myeloma. *Oral Surg Oral Med Oral Pathol* 1988;**65**:533—7.
2. Epstein JB, Voss NJS, Stevenson-Moore P. Maxillofacial manifestations of multiple myeloma. An unusual case and review of the literature. *Oral Surg* 1984;**57**:267—71.
3. Langlais RP, Langland OE, Nortje CJ. Diagnostic imaging of the jaws. first ed. Baltimore: Wiliams and Wilkins; 1995. pp. 423—7.

4. Lynch MA, Brightman VJ, Greenberg MS. Burket's oral medicine. Diagnosis and treatment. J.B. Lippincott Co.; 1994. p. 331, 382, 528, 529.
5. Kelly JJ, Kyle RA, Miles JM, OBrien PC, Dyck PJ. The spectrum of peripheral neuropathy in myeloma. *Neurology* 1981;**31**: 24—31;
Scutellari PN, Orzincelo C. Mandibular lesions in multiple myeloma. *Radiol Med* 1992;**83**:219—23.
6. Rooper AH, Gorson KC. Current concepts: neuropathies associated with paraproteinemia. *N Engl J Med* 1998;**338**:1601—7.
7. Wambach G, Engelkirchen R, Saborowski F. Therapy resistent burning tongue as an early symptom of small-cell brochial carcinomA. *Med Welt* 1979;**30**:993—5.
8. Ferguson JW, Burton JF. Clinical presentation of acoustic nerve neuroma in the oral and maxillofacial region. *Oral Surg Oral Med Oral Pathol* 1990;**69**:672—5.