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CASE REPORT

Large infiltration of chronic lymphocytic leukemia in the mandibular region

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Summary Orofacial complications of leukemia include lymphadenopathy, gingival bleeding, petechiae, ulceration, gingival enlargement and infections. Chronic lymphocytic leukemia (CLL) is the most prevalent type of adult lymphoproliferative disorder, but oral manifestations are uncommon. We describe a rare case of extensive mandibular infiltration in chronic lymphocytic leukemia.

Introduction

Chronic lymphocytic leukemia is the most common type of leukemia, affecting mainly elderly adults. Diagnosis usually is made in asymptomatic individuals, showing lymphocytosis of ''mature'' B-cells. It has an indolent response and a variable outcome. Most of the cases are incurable leading to death in a period of two to twenty years.[1,2] The main cause of death is bacterial infection, consequent to decreased levels of immunoglobulin. CLL can cause anemia, thrombocytopenia, lymphadenopathy, hepatomegaly, splenomegaly, fatigue, excessive sweating, loss of weight and appetite and flank pain. Eventually CLL can suffer transformation to prolymphocytic leukemia, or the patients develop concurrently a large cell lymphoma, characterizing Richter's syndrome.[2–4] Primary oral involvement by CLL is rare.[5–7] We describe a patient with CLL presenting extensive neoplastic B cell infiltration in the mandibular region.

Case report

A 56-year-old man with 4-year of B-CLL was referred to the Dentistry Section of the Hematology Center, UNICAMP, Brazil. He had been treated with

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seven courses of chlorambucil 0.15 mg/kg/7 days, five courses of 2CDA (0.09 mg/kg) and anti-fungal and anti-bacterial prophylaxis. From diagnosis to first dental visit he presented fatigue, profuse night sweats, loss of weight and appetite, episodes of fever, infection of the respiratory tract, generalized lymphadenomegaly and splenomegaly.

Extra-oral examination revealed cervical, submandibular and facial lymphadenomegaly. Intraorally it was noted both alveolar ridge and gingival enlargement. The patient had only five teeth, all poorly conserved. At this moment the patient presented hemoglobin level of 14.0 g/dL, white blood cell count of 16.3×10^3 cells/μL with 62.4% lymphocytes, and a platelet count of 67×10^3 cells/μL. Two teeth were extracted in different days under local anesthesia with antimicrobial cover.

Two months later the patient developed a large mass involving the right maxilla, submandibular and cervical areas. Intraoral examination revealed an extensive fleshy mass, measuring 5.0×4.5 cm, in the right alveolar mucosa, extending from central incisor to the first bicuspid area, covered by a reddish mucosa. The tongue was elevated as the mass extended to the sublingual region. Axial and coronal CT scan showed a heterogeneous and expansive mass with poorly defined borders. The lesion involved facial, pterigoideo, sternocleidomastoideo, lingual, and parapharyngeal muscles, as well as submandibular and parotid salivary glands, all of right side. No bone involvement was noted (Figs. 1 and 2).

An incisional biopsy of the intraoral mass revealed an infiltration of homogeneous, small, hyperchromatic lymphocytes (Fig. 3), positive for CD5 and CD20, confirming tissue infiltration by CLL. The patient was treated with fludarabine, mitoxantrone and dexamethasone (FND protocol),[8] and most of the mass disappeared after the first course. Further the disease progressed, also involving the left side of the face. New FND cycle and palliative radiotherapy in both sides of the cervicofacial regions was indicated, with reduction of the mass. The patient's condition gradually deteriorated over the next 10 months and he died due to infection.

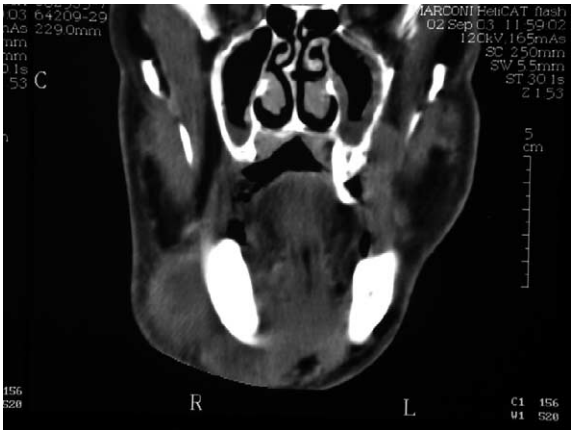

Figure 1 Computer tomography (axial projection) showing an expansive lesion of the right mandibula with poorly defined borders.

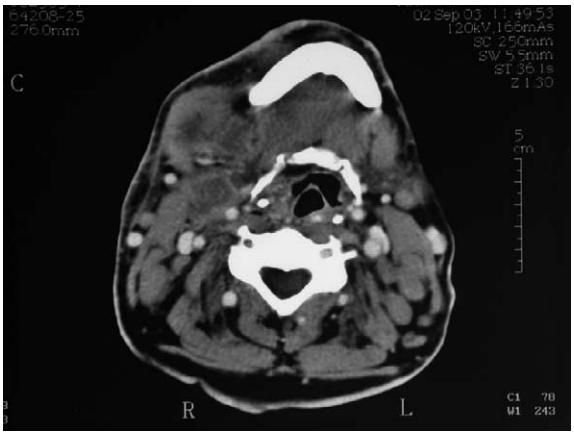

Figure 2 Computer tomography (coronal projection). The expansive tumor involves adjacent muscles and salivary glands.

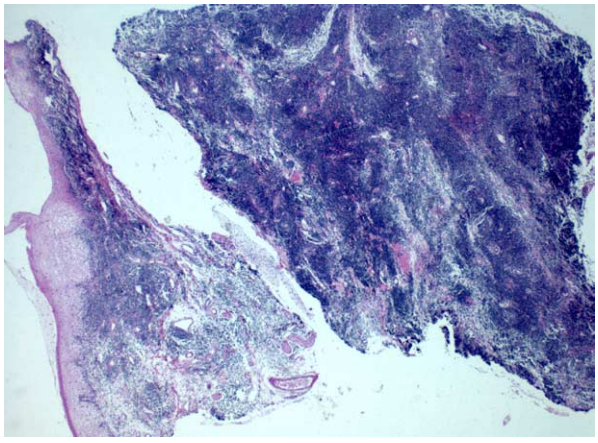

Figure 3 Biopsy of the intraoral mass showing that the tumor is formed by solid sheets of small, hyperchromatic, homogeneous lymphocytes (hematoxylin and eosin, original magnification ×25).

Discussion

Oral manifestations of leukemia are frequently described in acute leukemias, mainly of the myelocytic origin.[5,6,9] Few published papers report CLL involving the mouth. The oral manifestations described in CLL were gingival and palatal enlargement,

gingival bleeding, purpura, petechiae, ulceration, and pain.[6,7,10–12] Local factors as poorly oral hygiene and calculus also are associated with these oral manifestations in patients with leukemia.[6,7,12] Moreover, dental bacteria can be responsible for systemic infections specially in cases where immunodeficiency is associated with the disease and/or chemotherapy.

Therefore, multidisciplinary approach with dentist participation is very important for appropriated treatment of the patients with hematological malignances due to the large variety of oral manifestations and necessity of infectious focus removal. Moreover, the oral involvement may be the first sign of the disease and the earlier diagnosis is important for prognosis.

The patient here reported presented a large mass, and clinically it was considered the possibility of Richter's syndrome, but this was discarded by immunohistochemistry assay of the lesion. As far as we are aware, there are no previous reports of mass due to CLL infiltration into the mouth and it probably indicates a less favorable course of the disease.

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