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ORAL DISEASES Leading in Oral, Maxillofacial, Head & Neck Medicine WILEY

CLINICAL IMAGE

Gingival multilobulated growing-mass in HIV-positive patient

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KEYWORDS
Gingival mass, oral cancer, oral medicine, plasmacytoma, radiotherapy

1 | CASE REPORT

A 59-year-old man came to the emergency room of our department for the presence of a gingival mass that has lasted for 4 months. In his clinical history, the patient reported being affected by HIV since 1985 (currently treated with association of doravirine and abacavir/lamivudine); no further diseases were referred. No pain was reported by the patient and, at the clinical examination, no cervical lymphadenopathy or submandibular mass were observed. Intraorally, we could observe the presence of a single multi-lobulated mass, red in colour, localised at the buccal aspect of the incisal gingiva of the mandible. The lesion was painless, with elastic-soft consistency and had a longitudinal extension of approximately 1.5 cm (Figure 1a). Orthopantomography and periapical x-ray did not show any sign of bone involvement, thus an incisional biopsy was performed. Initial laboratory data showed: haemoglobin 16.4 g/dL; haematocrit 48.4%; white blood cell count 8100/mL; platelet count 188,000/mL; CD4 count 571/mm^3; CD8 count 2203/mm^3; HIV RNA not detected. Liver function tests revealed the following concentrations: albumin 4.27 g/dL; total protein 8.30 g/dL; total bilirubin 0.46 mg/dL; alanine transaminase 27 IU/L; aspartate transaminase 23 IU/L; serum lactate dehydrogenase 250 IU/L.

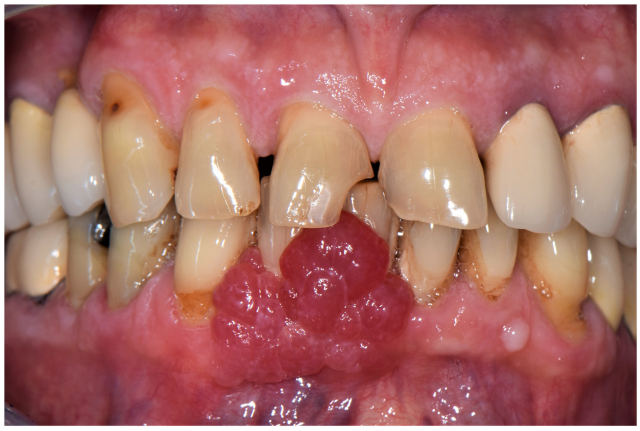

FIGURE 1 Multilobulated, painless gingival mass, red and purplish, without jaw-bone involvement.

2 | WHAT IS YOUR DIAGNOSIS?

Based on the patient's history, physical examination, and laboratory findings, which one of the following is the most suspicious diagnosis?

A Oral Kaposi's sarcoma
B Pyogenic granuloma
C Gingival extramedullary plasmacytoma
D Peripheral giant-cell granuloma

3 | DIAGNOSIS

The correct answer is C. In this case, our differential diagnosis of the gingival mass included Kaposi's sarcoma, pyogenic granuloma, peripheral giant cell granuloma and haematological and mesenchymal neoplasia (Lombardi et al., 2019, 2020).

The histopathological examination showed that the oral mucosa with stroma was entirely occupied by plasma cells with high-grade cytological atypia. The immunohistochemistry (IHC) showed positivity for CD138, MUM-1 and lambda, negativity for kappa, CD3 and CD20, while semiquantitative determination of Ki67 (MIB-1) was 90% (Figure 2). Further bone marrow biopsy, protein electrophoresis and urine lab-tests were negative for diagnosis of multiple myeloma, thus a definitive diagnosis of gingival extramedullary plasmacytoma was made. The positron emission tomography (PET) showed an enhanced up-take in the anterior area of the mandible ($SUV_{max} = 12$). The gingival lesion was treated by radiotherapy (RT) with a total dosage of 52 Gy divided in 27 fractions over a period of 40 days. After 6-months from RT, the complete remission of the lesion, without any sign of of recurrence, was observed (Figure 2).

Plasma cell neoplasms can be classified into three groups: multiple myeloma (MM), solitary bone plasmacytoma (SBP) (also known as medullary plasmacytoma) and extramedullary/extraosseous plasmacytoma (EMP) (Trivedi et al., 2016). EMP is defined as an extraosseous localised plasma cell neoplasm arising in tissues other than bone and which is characterised by a mass forming proliferation of monoclonal plasma cells, in the absence of underlying multiple myeloma (El-Naggar et al., 2017; Trivedi et al., 2016). EMP represents 5% of all plasma cell neoplasms and often involves head and neck area (Gonzalez-Perez & Borrero-Martin, 2016; Trivedi et al., 2016). Approximately 80% of EMP affects the upper respiratory tract: nasal cavity, paranasal sinuses, nasopharynx, oropharynx and larynx, while EMP occur less commonly in the gingiva (Gonzalez-Perez & Borrero-Martin, 2016; Trivedi et al., 2016). The median age at diagnosis of EMP is 55–60 years with a predilection for male patients (M:F ratio 3–4:1) (El-Naggar et al., 2017; Gonzalez-Perez &

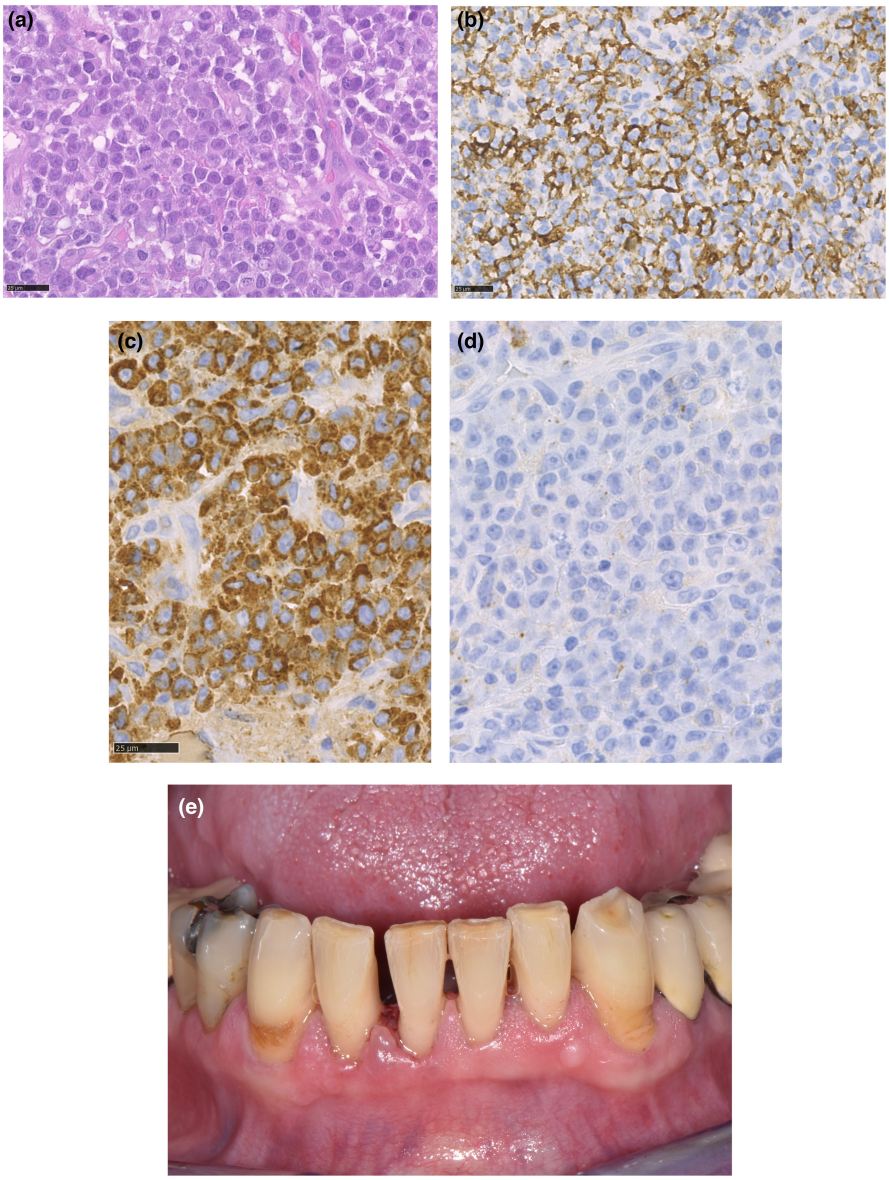

FIGURE 2 Histopathological examination revealed oral mucosa with stroma entirely occupied by plasma cells with high-grade cytological atypia, large eosinophilic nucleolus and high mitotic rate (a); immunohistochemical positivity for CD138 (b); immunohistochemical positivity for the lambda chain (c) and complete negativity for the kappa chain (d); clinical picture at 6 months after RT: complete remission of the lesion and absence of recurrence (e).

Borrero-Martin, 2016). EMP could be variable in size, ranging from one to several centimetres in diameters. Clinically, they usually appear as roundish, well defined, firm and sessile masses, but, in few cases, they even be multi-lobulated or pedunculated lesions showing local infiltration (De Camargo Moraes et al., 2016).

EMP is highly sensitive to radiotherapy (RT), which is considered the preferential treatment for localised lesions in 80%–100% of patients (Trivedi et al., 2016). The radiation dose should be in the range of 40–50 Gy (De Camargo Moraes et al., 2016). Approximately 70% of patients remains disease-free after 10 years from treatment, showing a better prognosis than those affected by SBP or MM (Gonzalez-Perez & Borrero-Martin, 2016; Trivedi et al., 2016).

After receiving RT for extramedullary plasmacytoma, a close follow-up is strongly recommended considering the risk of EMP conversion to MM, which has been estimated to range from 20% to 40% of cases (De Camargo Moraes et al., 2016; Gonzalez-Perez & Borrero-Martin, 2016; Trivedi et al., 2016).

At the best of our knowledge, there are only few cases of EMP affecting the oral cavity in HIV-positive patients, published in the literature (De Camargo Moraes et al., 2016). Persistent gingival masses are a common finding in daily clinical practice; incisional biopsy, histopathologic examination and immunohistochemistry are mandatory to achieve a definitive diagnosis and set up the most adequate treatment.

4 | OUTCOME

In cases of gingival masses, differential diagnosis should include inflammatory hyperplastic lesions, such as pyogenic granuloma or peripheral giant cell granuloma, but also malignant lesions, including extramedullary multiple myeloma, non-Hodgkin lymphoma, oral Kaposi's sarcoma and metastatic carcinoma (Gonzalez-Perez & Borrero-Martin, 2016; Lombardi et al., 2019, 2020). Even if rare, EMP is one of the possible diagnosis associated with persistent gingival mass lesions.

AUTHOR CONTRIBUTIONS

N.L. wrote the manuscript, collected the clinical data and followed the patient during diagnosis. E.V. writing review & editing, collected the clinical data and followed the patient during diagnosis. L.M. reviewed the manuscript and performed the histopathological diagnosis. G.L. reviewed the manuscript and followed the patient during diagnosis.

CONFLICT OF INTEREST STATEMENT

All authors have no conflicts of interest to disclose.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

PATIENT'S INFORMED CONSENT

The patient reported in this manuscript provided written informed consent for the publication of the case details.

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