Orofacial granulomatosis: do not forget leishmaniasis

Imen Boukhris, Samira Azzabi, Eya Cherif, Ines Kechaou

Department of Internal Medicine, Charles Nicolle's Hospital, Tunis, Tunisia

Correspondence to Dr Eya Cherif, eyacherif@yahoo.fr

Accepted 9 August 2015

DESCRIPTION

A 24-year-old man presented with a 1-year history of increasing swelling in the upper lip with a painful ulcer on the external oral mucosa. On physical examination, the upper lip showed swelling cheilitis, crusting and fissuring lesions. On the external fold of the upper lip, there were ulcerative lesions with necrotic and oedematous labial mucosa (figure 1A, B). The patient did not show any other clinical or biological abnormalities. Upper lip biopsy revealed organised non-caseating granulomas containing epithelioid cells and multinucleated giant cells surrounded by nodular inflammatory infiltrate composed of lymphocytes and plasma cells. A diagnosis of orofacial granulomatosis was made. Tuberculosis, sarcoidosis, Crohn's disease and lymphoma were eliminated as possible causes. Melkersson-Rosenthal syndrome and foreign body giant cell granuloma were also ruled out. Further microbiological investigations were carried out in order to locate the granulomatosis-causing agents. The serology for Leishmania spp was positive. PCR of an upper lip fragment confirmed the diagnosis of leishmaniasis. The patient was treated with intramuscular Glucantime for 20 days. The lesions steadily improved with total recovery (figure 2).

Proposed causes of orofacial granulomatosis include Crohn's disease, sarcoidosis, tuberculosis, foreign body giant cell granuloma and





Figure 1 (A) and (B) Profile view showing swelling cheilitis, crusting and fissuring lesions.



Figure 2 Evolution within a 12-week period.

Melkersson-Rosenthal syndrome. Other causes of granulomatous diseases must be investigated in order to find uncommon aetiologies. ¹ ² Mucocutanous leishmaniasis most commonly affects the upper respiratory tract. Oral mucosa involvement is uncommon. As seen in this case, the efficacy of meglumine antimoniate for treatment of leishmaniasis is established. ³

Learning points

- Clinician should bear in mind leishmaniasis in the differential diagnosis of orofacial granulomatosis.
- ► This case illustrates the need for an accurate and rapid diagnosis of leishmaniasis.
- Early diagnosis should prevent cosmetically disturbing cases.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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To cite: Boukhris I, Azzabi S, Cherif E, et al. BMJ Case Rep Published online: [please include Day Month Year] doi:10.1136/ bcr-2015-211919

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