What is your diagnosis?

Chronic ulcerated lesion of the nasal vestibule

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1. Clinical history

Mr Y, 26 years old, with no notable history, presented with bilateral nasal obstruction associated with rhinitis, mild epistaxis and an ulcerated lesion in the nasal vestibule, progressively invading the upper lip that had been present for almost 2 years (Fig. 1). Biopsy was performed in 2009 looking for a systemic disease or a site of infection, and histological examination concluded on chronic inflammation. In view of this inconclusive biopsy result, a laboratory work-up was performed to exclude syphilis, which gave negative results. A second biopsy was performed for histological and bacteriological examination. Histology revealed features of chronic inflammation, while bacteriology demonstrated Staphylococcus aureus infection with negative AFB tests. On the basis of these bacteriology results, the patient was treated with topical fusidic acid and systemic ampicillin.

Fig. 1. Lesion of the left nasal fossa invading the upper lip.

Fig. 2. Granulomatous inflammatory infiltrate with the presence of histiocytes containing numerous Leishmania bodies.

Due to absence of improvement in response to treatment, the patient consulted our unit, where another biopsy (skin and mucosa) was performed for histological and parasitological examination (Fig. 2).

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2. Answer

The diagnosis is primary nasal leishmaniasis, as histological examination revealed a granulomatous inflammatory infiltrate in the lamina propria with the presence of histiocytes containing numerous Leishmania bodies. Parasitological examination confirmed the diagnosis of nasal leishmaniasis: presence of Leishmania bodies inside macrophages.

A favourable course was observed in response to treatment with meglumine antimoniate at the dose of 20 mg/kg/day for 30 days with improvement of nasal symptoms and healing of the skin and mucosal lesion, and absence of parasites on follow-up biopsy 6 months after starting treatment.

3. Discussion

Leishmaniasis is a parasitic infestation classified by WHO among the leading public health problems in the world [1,2]. It can present in three forms: cutaneous, visceral and mucocutaneous [1].

Mucosal involvement during leishmaniasis is rare in the old world [3]. Most of the reported cases are observed in East Africa due to L. donovani and L. aethiopica infections. Only a few cases have been reported in Mediterranean countries, mostly in immunodepressed subjects [3]. However, rare cases of mucosal leishmaniasis in immunocompetent subjects have been reported in Spain; the zymodeme usually isolated was Leishmania infantum MON-1 [4].

Among the thousands of cases of leishmaniasis observed in Morocco, only two cases of mucocutaneous lesions have been reported [1]. We therefore report the third case of leishmaniasis of the nasal mucosa in view of the presence of nasal symptoms prior to onset of the skin lesion, the inconclusive findings on the first two skin biopsies and the absence of an adjacent skin scar.

In contrast with classically invasive and destructive new world mucocutaneous leishmaniasis, which is relatively refractory to conventional anti-leishmanial drugs, the mucosal lesion observed in this patient healed completely without causing any mutilation of deep tissues and responded favourably to meglumine derivatives with complete resolution after two months. Similar findings have been reported in other cases in Morocco, Tunisia and Algeria. These clinical data could suggest that the causal parasitic agent is not adapted to mucous membranes and is probably less virulent [1].

The parasitic species most frequently isolated in cases of mucocutaneous leishmaniasis are Leishmania braziliensis and Leishmania panamensis present in Central America. In two-thirds of cases, the mucosal site of parasitic infestation is due to haematogenous or lymphatic spread from a primary cutaneous site.

These two species have never been isolated in Mediterranean countries. However, culture on Novy-Neal-Nicolle medium identified the Leishmania zymodemes (MON) responsible for the cases of mucocutaneous leishmaniasis described in Tunisia. The species isolated were Leishmania major zymodeme MON5 and L. infantum zymodeme MON24 [3].

These two species are responsible for pure cutaneous forms of leishmaniasis in Morocco and in other Mediterranean countries [5]. These findings may reflect a change in the ecological profile and tropism of the parasite in the region, indicating the need for a large-scale national parasitological study in order to identify the serotypes and re-evaluate the status of Leishmania infestations in Morocco.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References