Leishmaniasis of the Lip

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Sir,

Leishmaniasis of the lips occurs mainly in young subjects (1–6). It is characterized clinically by the slow and progressive enlargement of one or both lips: macrocheilia is the final appearance (1–3, 5, 7). A nodule often present within the swelling undergoes an ulceration which may be covered by a crust (1, 2, 4–6). Bacterial superinfection is possible (5). The consistency of the entire lesion is parenchymatous-hard (2, 7). The swelling is often painful (4, 5). Patients with leishmaniasis of the lips are in good general health (1–3, 6); in particular, regional lymph nodes are never involved (1, 5–7). We present a rare case of cutaneous leishmaniasis on the lower lip in an Italian patient.

CASE REPORT

A 71-year-old man was admitted to our Institute because of swelling of the lower lip. The patient stated that he was in good general health and that he was not taking any medication. He also stated that the swelling had appeared approximately one year previously and that it had slowly enlarged to the current morphology and size. Two biopsies had previously been carried out at other hospitals, and in both cases a histopathological diagnosis of macrocheilits was made. The patient was not treated.

Dermatological examination showed the presence of a swelling that involved the entire lower lip. The surface
of the swelling was irregular; an oval ulcer localized on the right side of the lip was also present. The colour of the swelling ranged from pink to red (Fig. 1a). Consistency was parenchymatous-hard. The patient complained of pain. General physical examination did not show anything pathological; in particular, regional lymph nodes were not involved.

Laboratory and instrumental examinations were within normal ranges or negative. The patient was subjected to a new biopsy. A histopathological diagnosis of leishmaniasis was made (Fig. 2): this diagnosis was surprising, because the patient lived in Carrara (Tuscany, Mid-Western Italy), an area which is not endemic for leishmaniasis; moreover, the patient never travelled outside Italy. Cultural examination on Novy-MacNeal-Nicolle medium was positive for *Leishmania infantum*.

The patient was treated with i.m. N-methylglucamine antimonate (100 mg kg$^{-1}$ day$^{-1}$ for 2 weeks). We did not use this drug intra-lesionally because of the level of pain reported by the patient. The swelling decreased almost completely in approximately 1 month (Fig. 1b). No relapses were observed during follow-up (27 months).

**DISCUSSION**

In 1990, Sithéeque et al. (8) reported on a group of 492 patients with cutaneous leishmaniasis localized to the lips and peri-oral regions. Subsequently, El-Hoshy (1) published 12 cases of cutaneous leishmaniasis with localization on the lips, claiming that this is frequent although rarely reported in the literature. However, on the basis of the cases published so far (2–7), cutaneous leishmaniasis of the lips appears to be very rare outside Saudi Arabia. Furthermore, some cases described as leishmaniasis of the lips were actually examples of leishmaniasis of the face (4, 6).

In the rare cases in which it was possible to determine the responsible species, *Leishmania donovani* (7), *Leishmania major* (5) and *Leishmania tropica* (1, 2) were demonstrated. Clinical diagnosis of leishmaniasis of the lips is difficult: the most important diseases that must be taken into consideration in differential diagnosis are syphilitic chancre, cutaneous tuberculosis, granulomatous cheilitis, Melkersson-Rosenthal syndrome and squamous cell carcinoma.

As far as therapy is concerned, with the exception of a case of spontaneous complete remission (3) all the other patients were successfully treated with pentavalent antimony derivatives by the endovenous (6), intramuscular (1, 4, 5) or intralesional route (2, 7). No improvement was observed by means of cryotherapy or oral ketoconazole (1).

**REFERENCES**