Atypical Localization of Cutaneous Leishmaniasis

Sir,

Cutaneous leishmaniasis (CL) is a relatively frequent pathology in the Mediterranean basin and in Sardinia (Italy), where it has an incidence of approximately 0.16/1,000 inhabitants (1). In this region the disease is mostly due to *Leishmania infantum*, and vectors are *Phlebotomus perniciosus* in 80% of the cases and *Phlebotomus perfiliewi perfiliewi* in 20% (1). The main CL reservoir is represented by dogs but also rodents have been incriminated. Disease onset occurs after a 2–4 week incubation period and is generally characterised by the appearance of a single nodular lesion, preferentially on the face or forearm (2). We here report a recent case with an unusual morphology and disease location.

CASE REPORT

A 45-year-old man, a building contractor with hunting as a hobby, had a 5-month history of an erythemato-infiltrative and intensely pruritic lesion in the perianal region, which had been treated with antihaemorrhoids, antibiotics, antimycotics and corticosteroids, leading progressively to worsening of the objective and subjective symptomatology. Remote anamnesis indicated chronic hepatitis B virus, well-compensated at clinical and laboratory levels. At the time of referral we observed a large oval-shaped plaque in the perianal region, with a maximum diameter of 8 cm. The lesion was bright red to violaceous-red in colour, had a hard consistency and was badly demarcated due to perilesional oedema but not painful (Fig. 1). Routine laboratory tests revealed a slight increase of transaminase and gamma GT and moderate thrombocytopenia. Humoral and cell-mediated immunity was normal. Histologic examination showed a dense infiltrate below a mildly hyperkeratotic and acanthotic epidermis involving the skin and part of the hypoderm and consisting of lymphocytes, plasmocytes, rare neutrophils and histiocytes that were particularly abundant in the papillary dermis. In the cytoplasm of the latter numerous *Leishmania* were present and could also be observed in groups outside the cells. Protozoal typing made it possible to identify *Leishmania infantum Zymodeme Montpellier 111*. The determination of anti-*Leishmania* antibodies in serum was negative.

Chest X-rays and abdominal echography were within normal range. Also the search for visceral localization of *Leishmania* and bone marrow biopsy yielded negative results.

Therapy was started with a single oral dose of itraconazole 200 mg/day for 2 months but with no benefit whatsoever. In fact the clinical pattern was the same and control histology, performed 1 month after the end of therapy, showed no changes in the infiltrate

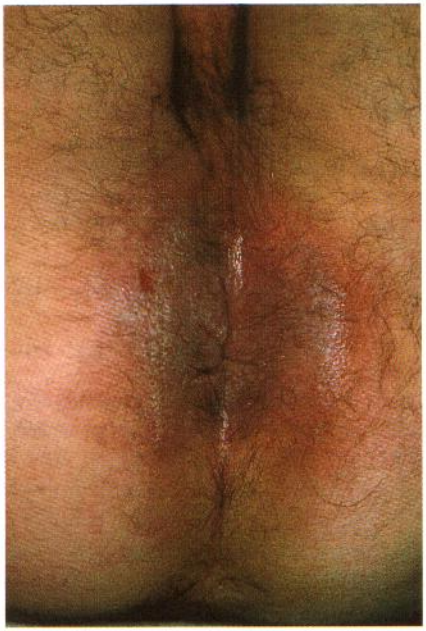

Fig. 1. Large oval-shaped plaque in perianal region.

or in the presence of *Leishmania*. A treatment cycle based on the administration of meglumine antimoniate at an intramuscular dose of 40 mg/kg/day was therefore started. After only 1 week of treatment there was a marked improvement of the clinical pattern, but on the 12th day the patient developed a diffuse and intense urticarial reaction, for which reason treatment had to be stopped. Control histology 30 days after the last dose still showed a considerable number of *Leishmania*. Consequently rifampicin was given orally at a dose of 600 mg/day for 2 months. At the end of this treatment cycle clinical recovery was almost complete, and histologic control 1 month later revealed a modest and aspecific infiltrate totally lacking *Leishmania*. After 6 months the clinical symptomatology was completely resolved and the search for parasites negative.

DISCUSSION

The perianal region is an extremely unusual site for CL, and to our knowledge this is the first case reported. As the protozoa is inoculated into the host by *Phlebotomus* we decided to look

for an explanation by studying the patient's habits. In fact, after several talks he told us that when he went hunting in Sardinia, where the disease has focal endemic areas, he was often bitten by *Phlebotomus* during defecation in the open.

In the diagnosis of leishmaniasis we often observe small nodules such as the "oriental sore". The atypical clinical pattern in our case may be due to the anatomical site of the lesion, a particularly favourable region for infiltration, and/or to pathomorphism as a consequence of the numerous and inappropriate topical treatments previously applied.

The therapeutical procedure normally applied by us in the cure of CL consists of intralesional infiltration with meglumine antimoniate and has allowed us to achieve excellent results without significant side-effects (3, 4). However, in this case, because of the site and extension of the lesion, the protozoal density and the disease duration, our preference was given to systemic therapy. Itraconazole, which has been reported by other authors as an active drug (5), was totally ineffective in our case. On the other hand, meglumine antimoniate triggered a severe urticarial rash, necessitating the interruption of treatment. The excellent results obtained with rifampicin without any clinical or laboratory adverse effects confirm the therapeutical value of this molecule (6). We therefore believe that it may represent a valuable alternative in the cure of extensive multifocal CL or in case of intolerance to meglumine antimoniate.

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