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 perfiliei* 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 erythematous-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 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, plasmaocytes, 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 specific *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 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...
Histiocytic Necrotizing Lymphadenitis (Kikuchi-Fujimoto's Disease) with Cutaneous Involvement

Sir,

Histiocytic necrotizing lymphadenitis (Kikuchi-Fujimoto's disease) was first described in 1972 as a benign lymphadenopathy with a self-limited clinical course. Later, this disease was frequently reported in Japan but rarely in Europe and in the United States. Its aetiology is unknown, possibly caused by a virus. The disease primarily affects cervical lymph node of young adult women, with fever and leukopenia. The histopathological features in the lymph nodes are peculiar; nodular areas with necrotizing foci in the cortex and paracortical areas of lymph node composed of foamy histiocytes and lymphoid cells, neutrophils being absent (1).

Cutaneous involvement is not frequent; when present, it is usually characterised by short and transient skin rashes (2). Some authors described shorter skin manifestations as papules (3, 4) or nodules (5) which showed similar histopathological features as the affected lesion in the lymph node.

An association with or a possible evolution of Kikuchi's disease to systemic lupus erythematosus has been previously described (6). This kind of possible evolution in the skin of Kikuchi's disease should be considered in differential diagnosis with cutaneous lymphoid neoplasia such as large cell lymphomas or Hodgkin's disease (2, 7).

A 21-year-old Italian woman came to our observation for a red-brown erythematous plaque measuring 2 x 1 cm, developed on the left cheek, 1 year before with slow and progressive enlargement. In the past 6 years she had been admitted three times to the Medicine Department of our hospital for recurrent episodes of lymphadenopathy associated with systemic symptoms such as fever and leukopenia. Each episode lasted for 1 month; the previous two had been characterised by right cervical lymphadenopathy and interpreted as due to viral infection, according to serological positivity of Epstein-Barr virus antibodies. She went to the third admission because of left cervical lymphadenopathy, and a diagnosis of Kikuchi's disease was made, based on histopathological and immunohistochemical features of the lymph node biopsy.

Laboratory examinations, including serum titers for infectious diseases, autoantibodies and bone marrow biopsy, showed leukopenia (white cell count of 2,510/cmm) but no other pathological alterations. The skin lesion which addressed the patient to our Department appeared shortly after the last episode of left cervical lymphadenopathy. No lymphadenomegaly and no systemic symptoms were present at the time of our observation, and haematological data were normal.

A total excision of the cutaneous lesion was performed, and the histological examination of the skin biopsy showed heavy lymphoid infiltration of the dermis and subcutaneous fat (Fig. 1). This lymphoid population did not show any epidermotropism and was mainly constituted by small round lymphocytes with scarce cytoplasm; scattered large mononuclear cells, with an irregularly shaped nucleus, could easily be seen. Among the lymphoid nodules, necrotic foci were present, surrounded by histiocytes with no granulocytes.

The immunohistochemical analysis of the lymphoid infiltrate failed to demonstrate any mononuclear populations. The mononuclear cells were positive for UCHL1/CD45 RO and CD1/CD43. In view of the morphological and immunophenotypical features, we consider this case as cutaneous involvement of Kikuchi's disease.

REFERENCES

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