Case report

Visceral leishmaniasis complicating systemic lupus erythematosus

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SUMMARY Active systemic lupus erythematosus in a 32-year-old Chinese woman was successfully controlled by plasmapheresis and steroids. However, occult visceral leishmaniasis was uncovered during therapy and responded to appropriate treatment.

Foreign travel and immigration can bring protozoal infections acquired abroad to countries where they are infrequently seen. These may be occult infections uncovered by immunosuppression, sometimes with fatal consequences.¹ This case of visceral leishmaniasis in a patient with systemic lupus erythematosus (SLE) illustrates these points.

Case report

A Chinese woman aged 32 from Hong Kong presented in October 1979 with a 2-month history of generalised arthralgia, facial rash, and swelling of the face and lower limbs. There had been no previous ill health, and she had last visited Hong Kong in 1976. Examination revealed a facial erythematous 'butterfly' rash, periorbital and facial oedema, pleural and pericardial effusions, and oedema of the legs. Investigations showed Hb 8.4 g/dl, leucocytes $3.1 \times 10^9/l$, platelets $66 \times 10^9/l$. Urine microscopy revealed cellular casts and red cells, urinary 24 hr protein 1.96 g/24 hr, serum creatinine 143 $\mu\text{mol/l}$, creatine clearance 36 ml/min, and serum albumin 23 g/l. Anti-nuclear factor (ANF) was positive 300 units, DNA binding 37% (NR<10%), C3 and C4 complement levels were reduced, and IgG and IgM circulating immune complexes were present. A diagnosis of SLE was established.

Treatment was commenced with plasmapheresis (a total of 3 exchanges) and prednisolone 60 mg daily for 10 days, the dosage being gradually reduced over the next month to a maintenance dose of 7.5 mg

daily. This achieved complete remission of the disease; the haematological findings, renal function, serum albumin, and complement levels became normal, and DNA binding fell to 4.7%.

In December 1979 and throughout the following year she complained repeatedly of sternal and epigastric pain. In January 1981 malaise, weight loss, and bimodal fever reaching 40°C also developed. Examination revealed anaemia, sternal tenderness, 2 cm hepatomegaly, and 7 cm splenomegaly.

Investigations disclosed Hb 8.1 g/dl, leucocytes $1.6 \times 10^9/l$ (45% neutrophils, 51% lymphocytes, 4% monocytes), platelets $96 \times 10^9/l$, erythrocyte sedimentation rate (ESR) 140 mm/h, weakly positive C reactive protein, and negative direct antiglobulin test. Serum protein electrophoresis showed polyclonal elevation in gammaglobulins, serum IgG being raised 27 g/l (normal range 9–18 g/l). Renal and hepatic function were normal. ANF 3 units, negative DNA binding, and normal complement levels indicated inactive SLE. Cultures for bacteria including *Mycobacterium tuberculosis*, viral titres, Mantoux test (1 in 1000), and chest x-ray proved negative. Abdominal x-rays, serum amylase, and gastroscopy were normal.

Bone marrow aspiration revealed a diagnosis of visceral leishmaniasis and showed a cellular marrow with a marked increase in macrophages, most containing Leishman-Donovan (LD) bodies (Fig. 1). Leishman fluorescent antibody titre was positive at 1/80.

Treatment with sodium stibogluconate 600 mg intramuscularly daily² induced rapid clinical improvement, with resolution of pain, fever, and hepatosplenomegaly. After 25 days' treatment the

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Fig. 1 Bone marrow aspirate: macrophage containing *Leishman-Donovan* bodies. (Haematoxylin and eosin, $\times 50$).

ESR and full blood and platelet counts were normal, no LD bodies were seen in bone marrow aspirate, but mild hepatorenal dysfunction had occurred. Treatment was therefore stopped. The patient has remained well without recurrence of leishmaniasis or SLE on 5 mg prednisolone daily.

Discussion

This patient neither had a previous history of visceral leishmaniasis nor had visited an endemic area for 5 years. Nevertheless occult infection had occurred. This phenomenon is well documented.³ The incubation period of visceral leishmaniasis varies from a few months to many years. During this time the parasite

multiplies in the reticuloendothelial system, including liver, spleen, and bone marrow. This accounts for the marked hepatosplenomegaly, pancytopenia, and probably the sternal pain, the latter antedating subsequent symptoms and signs by many months in this case. The exact nature of the immunological response evoked by the parasite is uncertain. The parasite multiplies unchecked despite the marked rise in IgG. This case illustrates immunosuppressive therapy uncovering occult leishmaniasis; appropriate therapy prevented a fatal outcome.

A febrile patient with SLE is a difficult clinical problem. The cause may be infection or exacerbation of disease activity. Fever, weight loss, pancytopenia, and splenomegaly are well documented in SLE. However, if the patient has visited an area endemic for visceral leishmaniasis, a diligent search for LD bodies may be necessary to exclude this disease.

Endemic areas are not confined to China and the tropics but include Southern France, Italy, Sicily, Spain, Malta, Greece, and Russia.⁴ The disease has recently reached epidemic proportions in India with resurgence of the sandfly vector *Phlebotomus argentipes* following a reduction in the antimalarial spraying programmes.⁵ Travel to a from these areas is common, and untreated visceral leishmaniasis has a high mortality. Therefore doctors should remain aware of this disease, particularly in their immunosuppressed patients.

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