Visceral leishmaniasis in a patient with systemic lupus erythematosus from Colombia, Latin America

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Dear Editor,

Visceral leishmaniasis (VL) is a prevalent condition in tropical and subtropical countries in Africa, Asia, Europe and the Americas [1, 2]. In the last, where VL is caused by *Leishmania chagasi/infantum*, Colombia is one of the countries still with endemic areas [3]. Systemic lupus erythematosus (SLE) and its long-term treatment are associated with increased risk of infection. The association of these diseases, although reported, is uncommon.

A 40-year-old man from Sampues, Sucre, Colombia, diagnosed seven years earlier with SLE, presented with a 15-days history of fever, abdominal pain and distension, general malaise, and coluria. The highlights on physical examination were pallor, palpable hepatosplenomegaly (12 cm below the bilateral rib borders) and low-grade fever (Figure 1). When diagnosed with SLE, ANA and anti-DNA antibodies were positive, anticardiolipine negative, with C3 elevated and C4 normal. Since there, he was treated with prednisolone. Blood tests at income showed pancytopenia, hyperproteinemia with hypoalbuminemia and polyclonal hypergammaglobulinemia (Table 1). Electrophoresis showed a polyclonal gamma curve. Abdominal sonography revealed hepatosplenomegaly (Figure 1). Microbiology investigation was negative for the most common pathogens, including tuberculosis. There were no signs of haematologic malignancy in the bone marrow smear, but it was positive for amastigotes (Figure 1). The patient was treated with miltefosine and pentamidine, and immunosuppression was adjusted. No ECG alterations were observed during treatment (serially performed). He showed rapid clinical improvement (no pancytopenia at day 23) (Table 1), and 2 months later he had no signs of disease.

The differential diagnosis in a patient with SLE presenting with fever and multisystemic manifestations includes tropical infectious diseases when living or returning from endemic areas [1, 4, 5]. The patient lived in an endemic area for leishmaniasis, and typical clinical and laboratory changes were all present, making this case highly educational, as other reported before [1]. The case highlights the importance of a patient's epidemiological background and how it can lead to the diagnosis and timely treatment of a rare disease [1, 3]. Since the first case of such association reported in UK in 1983 in a traveler, most of the cases have been reported in Europe [6,7] including one case in Turkey [8]; beyond that, a case in India [9]. So far, no cases have been reported in the Americas.

In Brazil, there has been a report of three cases of VL mimicking the symptoms of SLE [10]. In endemic areas for VL, the diagnosis or exacerbation of SLE may be problematic. However, there are some characteristics that could help to distinguish each other: massive splenomegaly is not a common sign in SLE (presented in our patient), arthritis is not a common clinical feature of VL,

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Table 1 - Clinical laboratory tests evolution of the patient.

Variable	*Laboratory reference Range, adults*	*Day 1*	*Day 7*	*Day 9*	*Day 19*	*Day 21*	*Day 23*
Haematocrit (%)	35-50	26	23	23	-	30	35
Haemoglobin (g/dL)	11.0-16.5	8.6	7.3	7.03	-	10.1	11.15
White blood cell count (per mm^3)	5.0-10	3,800	5,200	5,200	-	2,700	5,700
Granulocytes (%)	43-76	86	40	40	-	-	55
Lymphocytes (%)	17-48	14	50	50	-	-	44
Platelets	150,000-450,000	113,000	210,000	210,000	130,000	126,000	168,000
Reticulocyte	<1	3.3	1	1	-	-	1
AST (IU/L)	8-40	47	-	-	-	128	35
ALT (IU/L)	≤45	22	-	-	-	134	40
Alkaline phosphatase (IU/L)	20-140	460	-	-	276	891	140
Total protein (g/dL)	6-8.3	-	100	-	-	-	-
Albumine (%)	58-70	-	22.7	-	-	-	-
Gammaglobuline (%)	8-17	-	65.4	-	-	-	-
Alpha 1 (%)	1-2.8	-	2.7	-	-	-	-
Alpha 2 (%)	7-12	-	4.8	-	-	-	-
Beta (%)	9-14	-	4.4	-	-	-	-

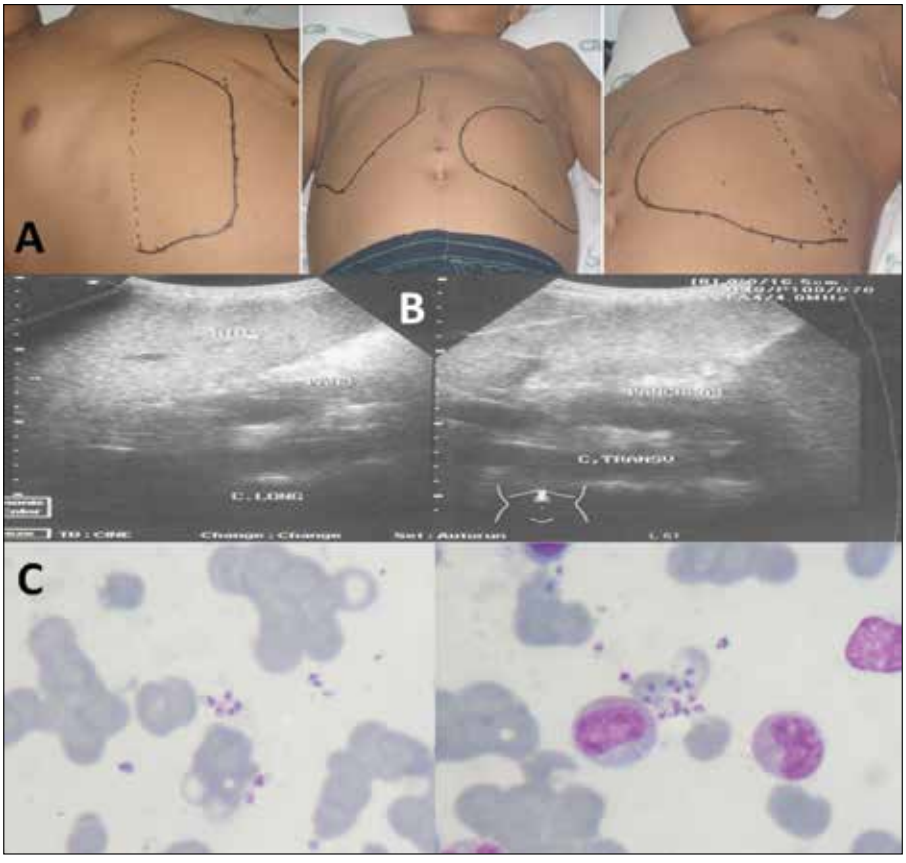

Figure 1 - Clinical findings. A) Hepatosplenomegaly B) Abdominal sonography showing hepatomegaly C) Bone marrow Giemsa-stained smear showing amastigotes.

high values of CRP are not common in SLE patients unless in the presence of infections, complement serum levels may be decreased during an SLE flare, but they usually are normal in VL, SLE patients with VL may present negative anti-*Leishmania* antibodies, but a positive test (at even low titers) should be valued. Given high fatality rate in cases of VL infection in SLE patients (up to 25%) prompt diagnosis and treatment is highly relevant. The combination of miltefosine and pentamidine has been suggested to be an appropriate treatment for cases of "chronic" VL that is sometimes observed in HIV-infected and other immunosuppressed patients [11].

This is a good opportunity to remember that VL may be a difficult task in immunocompromised subjects, such as SLE, due to a series of factors causing a drawback of cellular immunity, as it is also the case of HIV/AIDS or cirrhosis, that can favor parasites growth and disease manifestation [12, 13]. At the same time, in patients with chronic diseases, such as cirrhosis, VL can decompensate the patient [13]. Finally, it is also important to mention that VL may mimic a lupus flare and for that reason gold standard diagnoses for both are necessary. The clinical and laboratory features overlap in the two diseases [14].

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