

A novel case of human visceral leishmaniasis from the urban area of the city of Rio de Janeiro: autochthonous or imported from Spain ?

Rio de Janeiro, January 18, 2017

Dear editor

The State of Rio de Janeiro was considered free of human visceral leishmaniasis (VL) until the first case was detected in August 1977¹. Between 1977 and 2006 there were a total of 87 confirmed cases, all of which from peri-urban areas close to Pedra Branca and *Gericinó* Massifs in the West Zone of the city of Rio de Janeiro, Brazil¹. No new cases occurred in these areas since 2007². However, we have recently reported in this journal the case of a 29-year-old woman resident of *Cajú* neighborhood (central zone) who was diagnosed in January 2013 and whose case was considered by the Municipal Civil Defense and Health Surveillance to be the first instance of autochthonous VL in the urban area of the city of Rio de Janeiro³. We now wish to report a novel case of VL we had the opportunity to care for at the same ward of the same university hospital.

A 44-year-old previously healthy male patient was admitted in October 2016 with a one-year history of recurrent fever, anorexia, chronic fatigue, weight loss, increasing abdominal girth, epistaxis, and pedal edema. He sought medical advice on several occasions and at diverse facilities, but his illness remained undiagnosed. Symptomatic drugs were prescribed, as well as iron and cobalamin supplements to treat pancytopenia. His clinical status slowly progressed to a wasting disease.

The patient resided in *Brás de Pina* (North zone of the city of Rio de Janeiro) since August 2013, when he came back to Brazil after an uninterrupted stay of 23 years in Spain. He was a skilled mason and used to work for a Spanish construction company. In Spain, he lived mainly in the city of Almeria (southeast Mediterranean coast), but frequently traveled and established temporary residence throughout the country, including Madrid, Alicante, Seville and Barcelona. As the patient stated, he went “*anywhere they were building something*”. After returning to Brazil, he established himself in *Brás de Pina*, where he spent his days in the construction and improvement of his own house. He only left the city for a few days in December 2015 to spend Christmas time in his native town of Belo Horizonte, Minas Gerais State, Brazil.

Clinical examination revealed an undernourished patient, with pallor, tachycardia, tachypnea, hepatomegaly (10 cm from the right costal margin), splenomegaly (7 cm below the left costal margin), and ascites (Figure 1). Laboratory evaluations showed a hemoglobin of 8.6g/dL (13.8-17.2g/dL), a total white-cell count of 1,200 per mm³ (4,000-10,000 per mm³), a platelet count of 9.9 x 10⁴ per mm³ (15-45 x 10⁴ per mm³), an albumin of 1.7g/dL (3.5-5.2g/dL), and an activated partial thromboplastin time of 11.7 sec. (30-40 sec.). There was no serological evidence of hepatitis B, hepatitis C or HIV infection and no history of liver disease or alcohol abuse. A diagnosis of VL was made by demonstrating parasites consistent with *Leishmania* spp. at the amastigote stage of development in the bone marrow aspirate and the biopsy. Culture of a bone marrow aspirate yielded promastigote forms consistent with *Leishmania* spp. Treatment was initiated in October 13th, 2016, with a daily regimen of amphotericin B deoxycholate, starting with escalating doses, until a cumulative dose of 1g was reached. He experienced an uneventful recovery, was discharged four weeks later, and is being followed as an outpatient.

We found this case to be noteworthy since it could represent a new case of autochthonous VL in the urban area of the city of Rio de Janeiro. The patient's

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Received: 18 January 2017

Accepted: 20 January 2017

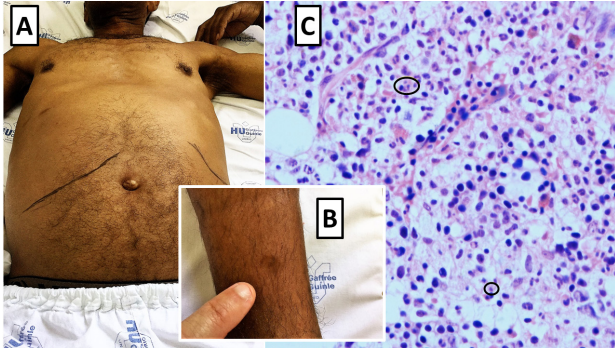


Figure 1 - Clinical images of a 44-year-old male patient diagnosed with VL in the urban area of the city of Rio de Janeiro. Marked hepatosplenomegaly as part of a wasting disease (A). Pitting edema in the anterior aspect of the left shin (B). Bone marrow biopsy stained by hematoxylin and eosin (C) shows a diffuse proliferation of histiocytes and intracellular forms (black circles) of *Leishmania* spp. amastigotes (original magnification 400 x).

house is located less than 10 km far from the neighborhood of *Cajú*, where our previous patient resided. We consider highly unlikely that he could have acquired this protozoal infection during the brief escape to celebrate Christmas in Belo Horizonte, since he was already symptomatic. Alternatively, we found it inevitable to raise the possibility of his infection having been acquired in Spain. In fact, Spain is experiencing an unprecedented outbreak of canine⁴ and human cutaneous and VL since 2009⁵. The disease is recorded nearly everywhere in the Peninsula and Balearic Islands, but seems to have hit more severely the outskirts of Madrid^{6,7}. A time span of 26 months separates his arrival in Rio de Janeiro and the start of his symptomatic disease. The incubation period of VL is in general, considered to be between two to eight months, and such a long interval may be unusual, but is not unknown. Descriptions of 24⁸, 34⁹, and at least 48 months¹⁰ between the exposure and the onset of disease have been reported. Moreover, an insidious onset of disease could have passed unnoticed.

We should constantly monitor and be prepared for changes in the epidemiology of the leishmaniasis. In Spain, both cutaneous and VL are caused by species *Leishmania* (*Leishmania*) *infantum*¹¹. Likewise, this species, also known as *Leishmania* (*Leishmania*) *chagasi*, is the etiological agent of VL in Brazil and in the Americas. In contrast, in the State of Rio de Janeiro, almost all cases of American tegumentary leishmaniasis (ATL) are caused by *Leishmania* (*Viannia*) *braziliensis*¹². It is remarkable that soon after our first report of autochthonous VL from an urban area of the city of Rio de Janeiro³, two cases of ATL were diagnosed from the same *Cajú* neighborhood¹³, but the species could not be identified. Interestingly, this was followed by the first report of ATL caused by *Leishmania* (*Leishmania*) *infantum* in an urban area of Rio de Janeiro¹⁴. The patient, an 81-year-old

woman, was also resident of *Cajú* neighborhood. All these observations followed the diagnosis of 25 cases of canine VL in *Cajú* neighborhood in 2011¹⁵.

At present, it remains unclear whether the patient reported herein represents a novel urban autochthonous case or if the disease was imported from Spain. Maybe future studies will help us clarify this issue. The former hypothesis should ring further alarm bells in surveillance and control programs. The latter reminds us that imported leishmaniasis should be regarded as a major travel medicine concern¹⁶ when travelling to endemic countries, and even to non-endemic ones if confirmed cases have already been reported. What is undisputable is that if not suspected and left untreated, VL has devastating consequences. The fact that our patient's illness remained undiagnosed for one year suggests that the awareness of the clinical spectrum of VL has waned. It is of great clinical and epidemiological importance that physicians be familiar with the presentation patterns of leishmaniasis, so that these diagnoses will not be missed or delayed.

Taken together, these cases of ATL and VL highlight the urgent need of surveillance and control programs in the city of Rio de Janeiro, including the active search of human and canine cases. The recent explosive Spanish outbreak should be taken as an alert to other areas where favorable eco-epidemiological conditions may exist.

CONFLICT OF INTEREST

The authors declare there is no conflict of interest.

INFORMED CONSENT

The informed consent of the patient was obtained for publication of the case.

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