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CASE REPORT

Diagnosis of *Leishmania* infection in a nonendemic area of South America

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American tegumentary leishmaniasis is the generic name for a variety of cutaneous and mucocutaneous presentations of parasitosis caused by several species of the genus *Leishmania*. This is a widespread infection in the American continent, from the South of the United States to the North of Argentina. We herein describe the management of a patient with American tegumentary leishmaniasis in Mendoza, Argentina, a nonendemic area of South America, whose diagnosis and treatment were significantly delayed, because the patient did not report a recent history of travel to any known endemic areas. This case stresses the need for training health-care professionals in the diagnosis and treatment of not only endemic parasitosis within their work zones but also nonendemic parasitosis.

Introduction

Leishmaniasis is a parasitic disease caused by protozoa of the genus *Leishmania*, which affects a large population all over the world. Estimates suggest that more than 12 million people are infected by the protozoan *Leishmania*. There are different species of *Leishmania* that can cause the disease in different regions. American tegumentary

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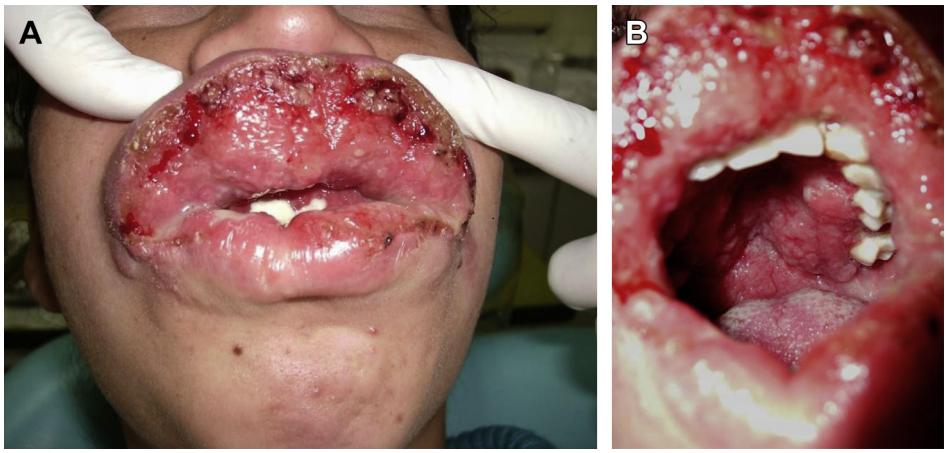

Figure 1. American tegumentary leishmaniasis. (A) Infection in the nose and lip regions that presented as edema and erythema. (B) Granulomatous lesion on the soft palate.

leishmaniasis is the generic name for a variety of cutaneous and mucocutaneous presentations of parasitosis caused by different species of *Leishmania*.[1] This infectious disease is a serious health problem. In endemic areas, high morbidity rates are reported, whereas in nonendemic areas it is usually misdiagnosed as some other similar granulomatous dermal diseases. Diagnosis of the infection is complicated because of the high costs of laboratory supplies and superinfection of the lesions. The main problem, however, is the insufficient training of health-care professionals in nonendemic areas.[2]

American tegumentary leishmaniasis is a widespread infection in the American continent, from the South of the United States to the North of Argentina.[3] The provinces of Salta, Jujuy, Tucumán, Catamarca, Santiago del Estero, Chaco, Formosa, Misiones, and Corrientes constitute the endemic area in Argentina. Although species of *Lutzomyia* (a vector of this disease) have been detected in the provinces of Santa Fe and Entre Ríos, so far there are no reports of infection in animals or humans.[4]

A common question asked by doctors whenever a patient presents with atypical clinical symptoms for the region is "Where have you been?" When investigating canine leishmaniasis cases, the veterinary surgeons usually inquire about the origin or movement of animals. A large proportion of leishmaniasis had been diagnosed in patients who either have been to or were born in an endemic area. Such questions should also be asked at the clinics of developing countries where nonautochthonous cases may appear.

In this report, we will describe the management of an American tegumentary leishmaniasis case that was detected in a nonendemic area. The diagnosis was significantly delayed because the patient did not report any recent history of travel to any known endemic areas.

Case report

Our case is a 19-year-old female patient who had never traveled outside the province of Mendoza, Argentina. As an antecedent to the present episode, at the age of 14, the patient suffered upper lip trauma while practicing sports, which caused edema and erosion in the affected area (i.e., the lips). At that time, she was diagnosed with *support granulomatous cheilitis*. She was treated with minocycline for a brief period in another institution, but the treatment was discontinued due to pharmacodermy. The patient mentioned that the lesions significantly increased in size during her two pregnancies, and that she did not receive any further treatment. In March 2013, she came to us for consultation because of severe granulomatous infiltration in the hard palate and soft upper and lower lips, which was associated with edema and mucosal erosions for 4 years (Fig. 1A and B). A nasal endoscopy showed granulomas in the nostrils, mouth, palate, and anterior and posterior pillars.

In April 2013, we performed a biopsy of the mucosa of the upper lip and the left posterior palate. Biopsy results showed intense inflammatory infiltrate with abundant lymphocytes, plasma cells with Russell bodies, and histiocytes with vacuolated cytoplasm. Rhinoscleroma was suspected.

Therefore, we requested additional tests to be performed on the patient: The patient's complete blood count was normal. The skin test for tuberculosis was negative. Her chest X-ray was normal. The venereal disease research laboratory result was negative (i.e., nonreactive). For the bacteriologic study, we collected mucus samples from the palate and lip. Neither alcohol-resistant acid bacillus nor fungal elements were observed in the sample. In addition, Giemsa and Ziehl—Neelsen staining were negative. Gram staining showed a few Gram-positive cocci. *Staphylococcus aureus* and *Streptococcus viridans* were isolated from the sample. She also underwent serology testing for deep mycoses. However, the mucus sample was negative for *Coccidioides*, *Paracoccidioides*, *Histoplasma*, and *Aspergillus*. The sample was negative for tumor markers as well.

In April 2013, despite failing to isolate *Klebsiella rhinoscleromatis* in culture, there was a clinical suspicion of rhinoscleroma based on biopsy results. The patient received ciprofloxacin (500 mg every 12 hours) for 4 months and cefixime (400 mg every 24 hours) for 1 month. A slight improvement in edema and erythema was observed in upper lip on the 1st month control visit. The patient remained stable on successive control visits.