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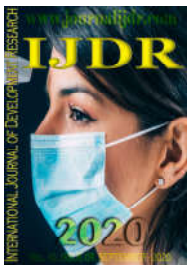

RESEARCH ARTICLE OPEN ACCESS

ORAL MANIFESTATION IN A MUCOCUTANEOUS LEISHMANIASIS CASE: A REPORT

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ABSTRACT

Cutaneous Leishmaniasis is a non-contagious, infectious-parasitic disease that can manifest itself in a mucocutaneous form, causing skin and oral mucosa lesions and compromising the patient's nutrition and life quality. The literature on leishmaniasis oral lesions is scarce, as the diagnosis is almost always based on nasal lesions, which represent 90% of MCL clinical manifestations. The present study aimed to report the case of a 78-year-old male patient diagnosed with MCL by a palate oral lesion biopsy.

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INTRODUCTION

Leishmaniasis in humans is a non-contagious infectious-parasitic disease caused by Leishmania genus protozoa, transmitted by infected Lutzomyia female sandflies bite (Brasil, 2017). Globally, is among the top ten neglected tropical diseases. Brazil is one of the ten countries in the world with the highest number of cutaneous leishmaniasis cases (Pan-American Health Organization, 2020). It is a disease widely dispersed throughout the world. In 2018, 92 countries or territories were considered endemic for and 83 had previously reported cases of cutaneous leishmaniasis (CL) and visceral leishmaniasis (VL) (WHO, 2018). In the Americas, leishmaniasis is endemic in 18 countries. The cases incidence in Brazil was considered very intense in the 2016-2018 triennium, according to the latest World Health Organization (WHO) report (PAHO, 2019). As it presents complex clinical manifestations and relevant epidemiological diversity, leishmaniasis is a serious public health issue, considered a compulsory notification disease 1. CL form can clinically manifest itself through cutaneous and/or mucous ulcerated lesions. Mucocutaneous leishmaniasis (MCL) presents more severe clinical manifestations, affecting places such as nose and mouth, which can lead to disfiguring scars and associated stigma (Bennis, 2018; Oyama, 2018). Oral lesions generally present as vegetative, granulomatous ulcers associated with painful symptoms, dysphagia and odynophagia and, consequently, compromised food intake, nutrition and the patient's life quality (Oliveira, 2013; Costa, 2014). The literature on leishmaniasis oral lesions diagnosis and treatment is scarce, as these are generally concomitant with nasal mucosa

Figure 1. Lesions on nasal and oral mucosa skin before (A, B and C) and after treatment (D, E and F)

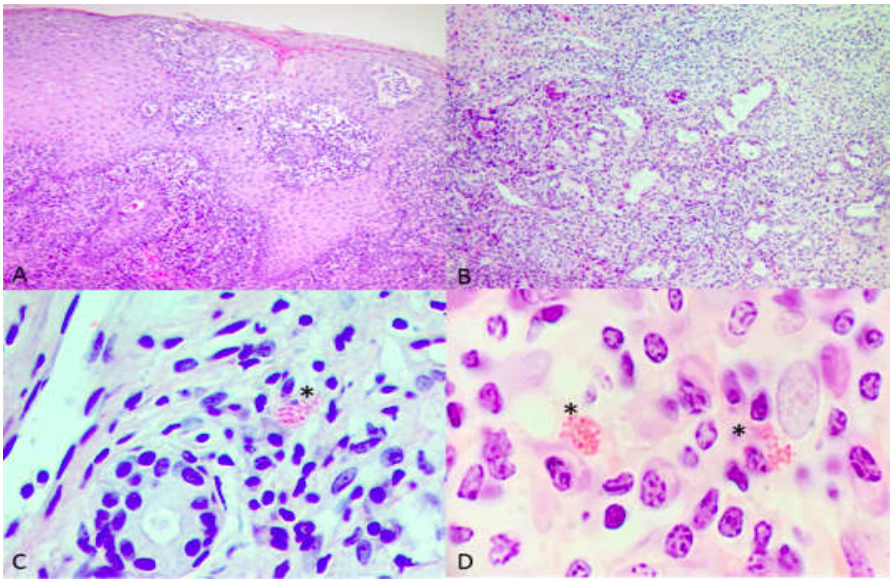

Figure 2. Pseudoepitheliomatous hyperplasia and diffuse intense chronic inflammatory infiltrate (A – 100X), replacement of accessory salivary gland tissue by the infiltrate (B – 100X). Small spherical eosinophilic structures, compatible with *Leishmania* sp. amastigote form were observed in macrophages cytoplasm (C – 400X and D- 1.000 X)

lesions, which represent 90% of MCL clinical manifestations and, in these cases, nasal biopsy is chosen to establish a diagnosis. It is reported a case of MCL which diagnosis was performed by examining a palate lesion.

CASE REPORT

A 78-year-old white male patient was referred to the Dental Specialties Center of the Unified Health System (SUS) in Ponta-Porã municipality, Mato Grosso do Sul state, Brazil, were he lived, due to a shapeless volume increase and erythema on the nose (fig.1- A), considerable asthenia and reported 40 kg weight lost, due to feeding difficulty. On intra-oral clinical examination, ulcerated, vegetative and granular-like lesions were observed in the hard palate region, soft palate, oropharynx and nasal cavity, with approximately one year evolution (fig.1- B and C). The initial diagnostic hypotheses were paracoccidioidomycosis and leishmaniasis. A palate incisional biopsy was indicated. The anatomopathological examination was performed by the Pathology discipline of the Mato Grosso do Sul Federal University (UFMS) Dentistry Faculty (FAODO) by the School of Dentistry (FO), University of São Paulo (USP) and revealed mucosa fragments coveredby stratified parakeratinized

epithelial tissue with pseudoepitheliomatous hyperplasia, ulceration areas, microabscesses, intense exocytosis and necrosis (Fig.2 – A and B). The lamina propria and submucosa connective tissue were practically replaced by an intense chronic inflammatory infiltrate, with a large amount of lymphocytes, plasmocytes and macrophages. Small spherical eosinophilic structures, compatible with Leishmania sp. amastigote form were observed in macrophages cytoplasm, confirming the diagnosis of Leishmaniasis (fig.2 – C and D). The patient was referred for treatment at DIA Hospital Professor Esterina Corsini, University Hospital (HU/UFMS). The treatment consisted of liposomal amphotericin B intravenous infusion in a hospital setting, considering the patient's advanced age and his history of cardiac arrhythmias and moderate renal dysfunction. The therapeutic protocol was 4 mg/kg/day daily doses until reaching the 3000 mg final dose. The patient returned for follow-up three months after treatment ended, showing considerable improvement: weight gain and remission of oral lesions. Healing areas and an important nasal deformity were identified (fig.1- D, E and F), but with no signs of recurrence. He was waiting for assistance to perform nasal plasty and subsequent prosthetic oral rehabilitation, remaining under monitoring for possible recurrence. This study was approved by the Mato Grosso do Sul Federal University Research Ethics Committee under opinion number: 3. 174. 741.

DISCUSSION

In 2018, Brazil reported the highest MCL case number (16.432 cases) in the Americas region, according to the World Health Organization (WHO) classification, with a 15.8/100.000 inhabitants incidence rate. Of all leshmaniasis cases reported, 265 were imported (1.6%), 95.1% were MCL cases and 17% of the total cases occurred at the borders4, as the case reported here. This may be due to the Brazilian geographical location, which borders ten countries, most terrestrially, then facilitating the spread of diseases and vectors9. As described in literature, as well as in the reported case, MCL lesions most frequently affect male patients, older than 10 years and appear after the resolution of a previous skin lesion or concomitantly with an active skin lesion (Costa, 2014; Gontijo, 2003). Cutaneous involvement is not a leishmaniasis pathognomonic sign11 and oral clinical manifestations can be very similar to those of paracoccidioidomycosis, tuberculosis, virchowian leprosy, tertiary syphilis, medium facial granuloma and some neoplasms (Oyama, 2018; Lessa, 2007) due to the granulomatous inflammatory reaction in the tissues common to these diseases. Thus, the diagnosis depends on the parasite identification associated or not with serological tests. In the present study, the histopathological evaluation allowed to observe the amastigote form of Leishmania sp. which, added to the oral and nasal lesions clinicalaspects, was sufficient for diagnosis conclusion. However, in some cases, the diagnosis becomes harder: infections secondary to injuries, high costs of complementary laboratory tests such as parasitological research and immunological diagnosis6, and, mainly, unqualified health professionals (Cargnelutti, 2016). The medication chosen for treatment was liposomal amphotericin B, due to the patient's advanced age, moderate renal function and cardiac arrhythmia. Despite the high cost, liposomal amphotericin B is efficient in all leishmaniasis forms treatment (Nicodemo, 2019). In this case, it was indicated as it is less nephrotoxic than conventional amphotericin B, in order to avoid worsening systemic conditions during treatment, which could require the therapy discontinuation (Hamill, 2013). MCL is the most disabling form of the disease. Its lesions can severely affect the patient's life quality by decreasing nutritional condition, consequent immune imbalance, deformities, and scars that remain even after treatment. The dentist role in this case is to promote early diagnosis, especially from oral cavity lesions, and prevent the disease systemic spread (Mignogna, 2015). Early diagnosis and correct referral for treatment are essential to determine a good disease prognosis and the patient's life quality, as they prevent disabilities and death, in addition to reducing the spread of the disease in the population, thus minimizing leishmaniasis consequences and public health impacts (WHO, 2014).

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