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ORAL DISEASES WILEY

CLINICAL IMAGE

Granulomatous ulcer affecting the hard and soft palate in a young patient

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1 | CASE REPORT

A 25-year-old male from a rural area, a non-smoker and non-drinker. He exhibits no systemic diseases and is not under continuous medication, but reported having been treated for cutaneous leishmaniasis at the age of 6. The patient reports the onset of painful lesions approximately 18 months ago, which occasionally bleed on the upper gum and soft palate.

During the physical examination, a granulomatous lesion was observed in the palatal region of the gum, extending from the left second molar to right canine. The lesion displayed painful sensitivity upon palpation, with all involved teeth exhibiting some degree of mobility. Another lesion was evident on the soft palate, characterized by poorly defined borders, resulting in uvula destruction and consequent loss of sensitivity in the affected area (Figure 1a,b). An incisional biopsy was performed under local anesthesia, and the specimen was sent for histopathological analysis. Additionally, culture for microorganisms was requested, as well as laboratory investigation for syphilis and polymerase chain reaction (PCR) for leishmania.

2 | WHAT IS YOUR DIAGNOSIS?

Based on the patient's history, physical examination, and laboratory findings, which one of the following is the most suspicious diagnosis?

A. Syphilis
B. Paracoccidioidomycosis
C. Aspergillosis
D. Leishmaniasis

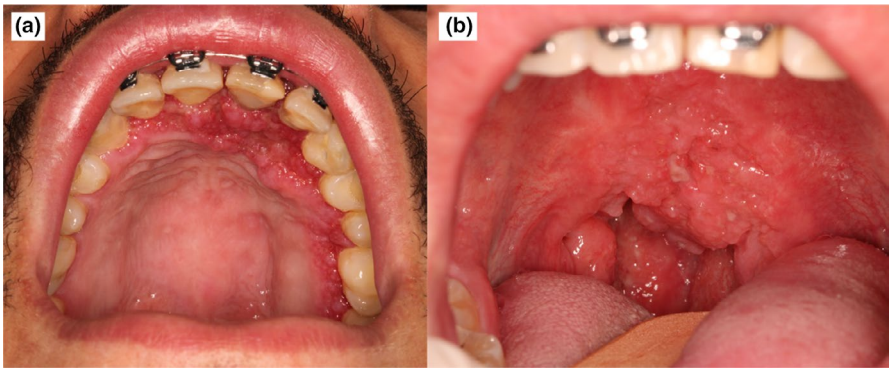

FIGURE 1 Clinical aspect showing slightly reddish moriform lesion in the palatal gum region (a) and diffuse lesion in the soft palate with a moriform and normochromic appearance resulting in destruction of the uvula (b).

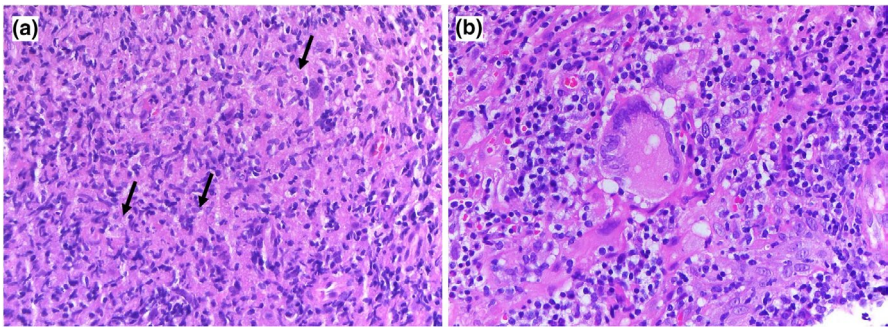

FIGURE 2 Microscopical aspect showing in (a) mononuclear chronic inflammatory infiltrate and a few cytoplastic inclusions compatible with microorganisms (black arrow) and (b) presence of multinucleated giant cells characteristic of granulomatous inflammation).

3 | DIAGNOSIS

The accurate diagnosis is D, leishmaniasis. Cultures for mycobacteria and fungi yielded negative results. Furthermore, the syphilis test returned negative. The biopsy conducted revealed a granulomatous inflammatory displaying a chronic inflammatory process (Figure 2a,b). Staining with periodic acid–Schiff and Ziehl-Neelsen stains produced negative outcomes. Therefore, the polymerase chain reaction (PCR) test confirmed the presence of Leishmania.

Leishmaniasis, caused by various Leishmania protozoan species, is a globally distributed parasitic disease (Mignogna et al., 2015). Brazil has a high incidence rate for all its forms, with transmission of the parasite occurs via insects of the genus *Phlebotomus* spp. or *Lutzomyia* spp. (dos Santos et al., 2020; Gontijo & Melo, 2004). The parasite has a dimorphic life cycle, with promastigote forms in the insect host and amastigote forms in host macrophages (dos Santos et al., 2020; Gontijo & Melo, 2004).

In humans, the clinical forms of the leishmaniases are broadly categorized into three groups: cutaneous (CL), mucocutaneous (MCL), and visceral leishmaniasis (VL) (Mignogna et al., 2015). In MCL, the primary cutaneous lesions are similar to the CL lesions; however, after various time frames, mucosal involvement may occur (Nadler et al., 2014).

Mucosal involvement is relatively rare, and the average duration until the onset of symptoms in cases of mucosal leishmaniasis (ML) is roughly 8 months following exposure or cutaneous lesion, with some instances extending up to 20 years after an untreated lesion or exposure (Mignogna et al., 2015). This may be attributed to hematogenous or lymphatic dissemination from cutaneous lesions, leading to parasitic infection and replication in naso-oropharyngeal mucosa macrophages, resulting in destructive inflammation (Talebi-Talghian & Ghogomu, 2023). This could potentially explain our patient's condition, as they mentioned being diagnosed with cutaneous leishmaniasis at the age of 6, with oral mucosa lesions appearing just two decades later.

Oral and perioral involvement can be atypical and potentially misleading, posing a diagnostic challenge for healthcare professionals. The differential diagnosis may encompass reactive lesions, infectious diseases such as bacterial (tuberculosis, tertiary syphilis), deep fungal infections, pemphigus vulgaris, pemphigoid, plasmacytic gingivitis, granulomatous diseases (including sarcoidosis and granulomatosis with polyangiitis), foreign bodies, and oral squamous cell carcinoma (Mignogna et al., 2015; Nadler et al., 2014; Pavli & Maltezou, 2010).

When of ML it affects the oral region, the posterior portion of the palate and the tongue are more commonly involved, although lip involvement has also been reported (Mignogna et al., 2015). The most common clinical presentation in the head and neck region is an exophytic lesion (Mignogna et al., 2015).

Diagnosing Leishmaniasis poses a significant challenge due to its diverse clinical manifestations, often mimicking other medical conditions. Timely identification of Leishmaniasis is crucial for minimizing the risk of mucocutaneous complications, including disfigurement and infection recurrence. As such, healthcare providers should include mucosal Leishmaniasis in the list of differential diagnoses for mucosal lesions.

4 | OUTCOME

The patient received treatment proposal based on use of 12 doses of liposomal amphotericin B 50 mg. After 10 doses, the patient had a complete clinical remission and his treatment was interrupted.

AUTHOR CONTRIBUTIONS

Isabel Schausltz Pereira Faustino: Conceptualization; writing – original draft; writing – review and editing. **Leonardo dos Santos Bayeh:** Investigation; formal analysis; writing – review and editing. **Danillo Batista Silveira:** Investigation; formal analysis; writing – review and editing. **Irineu Maia:** Investigation; writing – review and editing; formal analysis. **Joab Cabral Ramos:** Conceptualization; formal analysis; investigation; writing – review and editing; writing – original draft.

CONFLICT OF INTEREST STATEMENT

All authors have no conflicts of interest to disclose.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

PATIENT CONSENT

The patient reported in this manuscript provided written informed consent for the publication of the case details.

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