

## A rare case of leishmaniasis initially presenting with an erythematous nose

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### Abstract

**Background:** Leishmaniasis is a rare entity in the daily practice of an otolaryngologist, and its diagnosis can be challenging, resulting in delayed treatment. A broad spectrum of clinical manifestations depends on the species involved and the host's immune response.

**Case report:** We present an unusual case of a 90-year-old patient with progressively worsening erythematous, painful, and oedematous nose presenting a diagnostic dilemma.

**Conclusion:** Although leishmaniasis is uncommon, clinical suspicion should be raised in the presence of a relevant patient's history and certain clinical signs such as nasal and/or oropharyngeal lesions. HIPPOKRATIA 2025, 29 (1):32-34.

**Keywords:** Leishmaniasis, rhinophyma, rosacea, nose, diagnosis

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### Introduction

Leishmaniasis is a disease caused by protozoan parasites transmitted to humans by sandflies. Depending on the species involved and the host's immune response, a broad spectrum of clinical manifestations are seen including cutaneous, mucocutaneous, and visceral leishmaniasis (kala-azar)<sup>1</sup>. Nasal involvement in leishmaniasis is uncommon and can mimic other infectious or inflammatory diseases or malignancies, resulting in delayed diagnosis and management. Hereby, we outline an unusual case of a patient with progressively worsening erythematous, painful, and oedematous nose presenting a diagnostic dilemma.

### Description of the case

A 90-year-old patient presented to the Ear, Nose, and Throat (ENT) Department of the University Hospital of Alexandroupolis with a one-month history of progressively worsening nasal skin erythema, accompanied by swelling and pain, despite a prolonged course of oral antibiotics. The patient also complained of yellow nasal discharge, fever, and diffuse upper and lower mandible pain. There was no relevant pathological history.

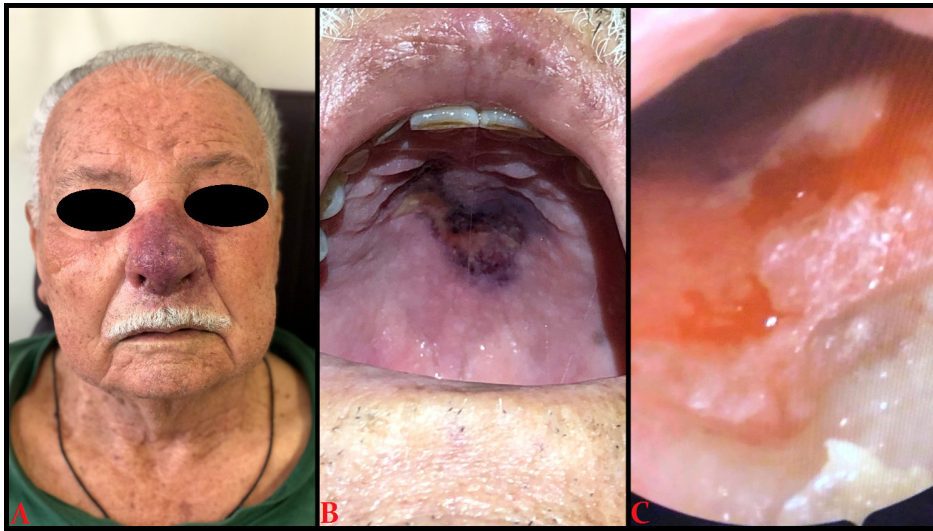
After the initial assessment by a dermatologist, the diagnosis of infected rhinophyma was made. ENT clinical examination confirmed the presence of nasal skin erythema, painful on palpation (Figure 1A). Interestingly, oral cavity evaluation revealed a dark-colored lesion on the anterior part of the hard palate (Figure 1B). On nasal en-

doscopy, mucosal edema with abundant nasal secretions was noted (Figure 1C). There were no elevated levels of inflammatory markers in blood testing. Computer tomography showed no bony destruction.

Cultures of nasal secretions were taken, and three different microorganisms were isolated (*Candida parapsilosis*, *Klebsiella pneumoniae*, and *Pseudomonas aeruginosa*); thus, targeted therapy with intravenous antibiotics and antifungals was commenced. In addition, biopsies from both nasal and palatal lesions were performed under local anesthesia. Despite the above medical therapy, the patient's condition continued to worsen. The histopathological features of the nasal specimen were indicative of rosacea acne. In contrast, the findings of the palatal biopsy raised a high suspicion of leishmaniasis with numerous tiny parasites present, although an obvious Leishman-Donovan body was not identified. Additional serological blood tests showed positive results for leishmania (IgG/IgM index of 2.8 with normal range <0.9). Unfortunately, further evaluation using advanced diagnostic techniques was not feasible in our institution.

Despite the absence of a definitive diagnosis and considering the above findings, the patient immediately started treatment with intravenous Amphotericin B (3 mg/kg/day for 10 days), with significant clinical improvement within a few days. Complete resolution of pain and swelling with disappearance of both nasal and palatal lesions was noted after two weeks (Figure 2). There was no recurrence at the 3-month follow-up.

### Discussion



**Figure 1:** Composite figure showing A) extensive nasal skin erythema, B) a hard palate lesion, and C) an endoscopic image of nasal mucosa edema with secretions.



**Figure 2:** Composite figure showing complete resolution of both lesions A) at the nose and B) at the hard palate, two weeks after initiation of treatment with Amphotericin B.

Mucocutaneous leishmaniasis is characterized by destructive granulomatous mucosal lesions affecting the nose, mouth, and pharynx. It is more common in immunocompromised patients and can occur if cutaneous leishmaniasis remains untreated or inadequately treated for a long time. In terms of immunological aspects, mucocutaneous leishmaniasis appears to be caused by excessive delayed type-1 hypersensitivity (DTH), leading to an increase in activated CD+4 T cells and a high expression of tumor necrosis factor (TNF) and interferon-gamma (IFN- $\gamma$ )<sup>2</sup>.

Laboratory criteria for the diagnosis of leishmaniasis are a positive serological test for antibody detection and the finding of the parasite (Leishman-Donovan bodies) in a clinical sample obtained by biopsy, punch, or scraping of lesion fragments. More specific methods include in vitro cultivation, polymerase chain reaction (PCR)-based methods, the Leishmania skin test (also known as the Montenegro skin test), and DNA sequencing<sup>3,4</sup>. Considering the fact that leishmaniasis is a rare disease with often

a non-specific presentation, the suspicion of its diagnosis by clinicians can be difficult<sup>5</sup>. Furthermore, the existing limitations of the diagnostic modalities available in a limited resource setting can make a definitive diagnosis even more challenging, resulting in a significant delay between presentation and successful diagnosis and therapy of the disease. In such cases, the initial diagnosis should be based on a clinical picture so empirical therapy can be given. A simultaneous infection can also be present, making diagnosis and management more complicated, as in our case.

Primary treatment options include systemic therapy with antifungals, antimonials, and/or immunomodulators<sup>6</sup>. Local therapies such as intralesional antimonials, topical paromomycin, thermotherapy, cryotherapy, and laser therapy have also been recommended as an alternative or in combination with systemic drugs in selected patients<sup>1</sup>. Close monitoring to check the patient's response to therapy is vital so prompt adjustments to the management plan can be made if required.

In conclusion, although leishmaniasis is not common,

physicians should be aware of the possibility of leishmaniasis in the presence of specific clinical signs such as nasal and/or oropharyngeal lesions, even in the absence of relevant travel history in order to avoid delayed diagnosis and management.

**Conflict of interest**

The authors declare no potential conflicts of interest concerning this article's research, authorship, and/or publication.

**Acknowledgment**

Written informed consent was obtained from the patient's legally authorized representative for anonymized patient information and images to be published in this article.

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