

A 48-Year-Old Man with Laryngeal Mass and Vocal Cord Palsy

(See page 1635 for the Photo Quiz.)

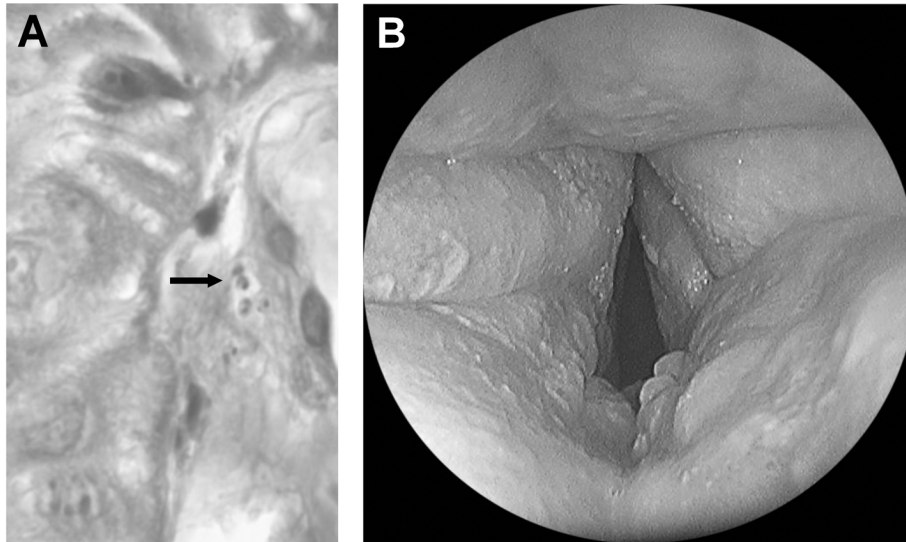


Figure 1. A, Histological examination of oropharyngeal biopsy specimen (May-Grünwald Giemsa staining; original magnification, $\times 1000$) showing intracellular amastigotes of *Leishmania* species (arrow). B, Laryngoscopic examination showing oropharyngeal area 3 months after completion of the treatment.

Diagnosis: mucosal leishmaniasis.

Examination of biopsy specimens failed to identify tumoral cells and revealed the presence of intracellular parasites that were suggestive of *Leishmania* amastigotes (Figure 1A). Serological test results were positive for *Leishmania* species, with a titer of 1:1280 ($n < 1:80$) in the blood. Results of quantitative, specific *Leishmania* polymerase chain reaction (PCR) performed on the laryngeal lesion specimen were strongly positive, with 400,000 parasites per million cells of tissue, but results were negative when PCR was performed on plasma samples.

The patient was treated with a 28-day course of meglumine antimoniate, which was administered at a dosage of 20 mg/kg intravenously. Tolerance was excellent, and the patient's dyspnea improved rapidly. The vocal cord palsy recovered completely after 6 weeks. Microlaryngoscopy at 3 months showed a regression of the laryngeal lesions (Figure 1B).

Leishmaniasis is a parasitic disease that is prevalent worldwide, is transmitted by the bite of infected female phlebotomus, and affects humans and some other vertebrates, such as

dogs and rats [1–5]. An estimated 12 million people are affected worldwide, with 2 million new cases each year [4]. The incidence of leishmaniasis has increased in settings with a high prevalence of human immunodeficiency virus (HIV) infection. Leishmaniasis is often considered to be an opportunistic infection [6–9].

Leishmaniasis has diverse clinical presentations related to the parasite species. Mucosal involvement of the nose, oral cavity, pharynx, or larynx occurs in 2%–5% of persons infected with *Leishmania braziliensis*. Generally, mucosal disease occurs 1 month to 2 decades after an initial cutaneous lesion.

Leishmaniasis presentations in patients who are co-infected with HIV are more likely to involve atypical locations, disseminated infection, and higher severity, especially among patients with a CD4⁺ cell count < 200 cells/mm³. In HIV-infected patients, leishmaniasis slows immune restoration and is associated with a high tendency to experience relapse [8–10]. Cases of leishmaniasis mimicking malignant disorders or developing in patients who have received a diagnosis of cancer have been

described [11–15]. This can either lead to misdiagnosis or cause a delay in the establishment of the correct diagnosis. Therefore, leishmaniasis should be considered in the differential diagnosis of malignancies in geographic areas in which it is endemic and in patients with a history of travel to these areas.

Our patient was probably infected while living in Portugal (where leishmaniasis is endemic) or during his annual trip there. The patient's high CD4⁺ cell count may explain the lack of general signs of the disease. However, local tissue damage as a result of radiotherapy could explain the spread and the high local viral load of parasites. Few cases of leishmaniasis after radiotherapy have been reported [14, 16].

There are several effective treatments for leishmaniasis. However, associated toxicities make them suboptimal. Pentavalent antimonials are recommended as first-line treatment for most patients, but they are associated with a high rate of primary resistance [17]. There are no data that establish mucosal leishmaniasis cure rates; however, there is evidence that pentavalent antimony, pentamidine, and amphotericin B may be equally effective [18]. Although current chemotherapy options result in a clinical cure, they seldom lead to parasitologic cure. Relapses usually occur within 6 months after treatment and are common among HIV-infected patients. In addition to anti-leishmanial chemotherapy, HIV-infected patients should receive antiretroviral therapy.

This case highlights the importance of histological diagnosis of mucosal lesions in patients with a medical history of oropharyngeal carcinoma. Histological confirmation of a suspected tumor resurgence should be standard practice.

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