Isolated laryngeal leishmaniasis in an immunocompetent patient: a case report

Un caso clinico di leishmaniosi primitiva laringea in paziente immunocompetente

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INTRODUCTION

Leishmania infantum spp is widespread in the world and has both visceral and cutaneous forms; the infection is endemic in the Mediterranean basin (southern and Tyrrhenian Italy), where the main animal reservoir is represented by dogs and the insect vector is the sandfly Phlebotomus spp [1, 2]. However, Leishmania infantum spp can sporadically cause an isolated laryngeal localization in a few patients living in endemic areas, who show risk factors for decreased immune response [3-5]. We describe a case of isolated laryngeal leishmaniasis randomly discovered in an adult Italian patient without current risk factors.

CASE REPORT

In December 2006, a 64-year-old male Italian resident of Naples was admitted to the Otolaryngology Department of the University of Naples “Federico II” due to dysphonia that had slowly increased in the previous 5-6 months. He did not complain of hoarseness, dysphagia, wheeziness or fever; he had a well-compensated type 2 diabetes mellitus (serum glucose level above 120 mg/dL); until 8 years before he had been a heavy smoker (50 cigarettes daily). Upon hospitalization, physical examinations and routine biochemical evaluations were normal, except for a slightly increased serum glucose level; an ultrasound scan of the abdomen showed no

Figure 1 - Direct laryngoscopy shows the presence of a lesion on the upper surface of left vocal cord (1A); after therapy for leishmania infection, the lesion significantly decreased (1B).
enlargement of the liver or spleen. A CT-scan revealed inhomogeneous thickening of the left wall of the pharynx with hyperplasia of the submandibular and laterocervical lymph nodes. As direct laryngoscopy showed a lesion on the upper surface of the left vocal cord (Figure 1A), the patient underwent a biopsy; histological examination (planned to detect a neoplasia) excluded the presence of neoplastic cells. Instead, oval/round shapes were found in histiocytes, consistent with *Leishmania* spp. amastigotes (Figures 2A-2B).

The patient was therefore referred to the Infectious Diseases Unit. Physical examination was still negative (absence of skin lesion, liver and spleen enlargement); abdominal ultrasound scan was again unremarkable. The antibody titre against *Leishmania* was weakly positive; the patient tested negative to antibodies against HIV, and CD4+ and CD8+ lymphocytes counts were normal. The patient underwent a bone marrow aspiration; both the histological examination and cultural growth were negative for *Leishmania infantum* spp. Thus a diagnosis of isolated laryngeal leishmaniasis was made; the patient was treated with liposomal amphotericin B, starting with a low dosage (0.5 mg/kg body weight) due to early signs of renal impairment (increased blood urea and creatinine levels); an overall dosage of 950 mg of liposomal amphotericin B was reached without other side effects. Two months later, the patient again underwent a laryngoscopy that showed a decreased lesion on the left vocal cord (Figure 1B); histological exam of the larynx biopsy was negative for haemoparasites. Laryngeal specimens were collected and sent to the Higher Institute of Health (ISS) in Rome for a DNA-PCR of *Leishmania*; this test was also negative (courtesy of Dr. Gramiccia).

**DISCUSSION**

In recent years an increase in visceral leishmaniasis has been seen in patients with immune compromission, namely patients with chronic disease (e.g. diabetes mellitus, asthma), with neoplasia or HIV infection, and in patients undergoing organ transplantation or immunosuppressive therapy [1, 6]. In this context, there appear to be few cases of isolated mucosal (and laryngeal) involvement by *Leishmania infantum*, which occurs in patients who are active smokers or who are treated by steroids due to chronic respiratory disease [3-5].

In contrast, in the case reported herein laryngeal leishmaniasis occurred in an immunocompetent patient who had no current risk factors: no active smoking in the previous eight-year period, absence of chronic obstructive respiratory disease and/or steroid and/or immunosuppressive treatment, no HIV infection and no decompensated diabetes mellitus. A past history of heavy smoking may well have caused laryngeal mucosal damage which could have predisposed the patient to local leishmanial infection. Hence heavy smokers, even if they have given up their habit, may have to be considered at risk for many years after. Since the clinical picture of laryngeal leishmaniasis is poor and the pathology can mimic many inflammatory or neoplastic diseases, we wonder how many patients, living or travelling in endemic areas, could have a mucosal infection that...
eludes correct diagnosis. Many patients are referred to the otolaryngology ward because of the onset of non-specific dysphonia; the leishmanial infection could be investigated in patients also without current risk factors. In such cases, it could be useful to collect fresh specimens for detection of protozoal DNA by PCR, since the latter test has been reported to have higher sensitivity than histological analysis [1, 3].

Finally, the problem could become more serious in the near future and involve countries usually considered not endemic for leishmaniasis. Indeed, the impact of climate change on the endemicity of infection in Italy and other parts of Europe has been recently reported [2]. In 2007, the winter was mild in the Mediterranean basin and also in the rest of Europe; spring and summer were also warm and experienced low rainfall. Such meteorological phenomena may well have led to an increase in the number of insect vectors and thus a recrudescence of Leishmania infantum infection in Europe. All physicians should be ready to suspect Leishmania spp as a possible culprit of non-specific laryngeal symptoms in patients living in Italy and other European countries, albeit immunocompetent and with no major risk factors.

Key words: leishmaniasis, larynx, immunocompetence.

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SUMMARY

Visceral leishmaniasis, a protozoan disease caused by Leishmania infantum, is endemic in the Mediterranean basin, especially southern and Tyrrhenian Italy. Its aetiological agent can also sporadically cause isolated laryngeal localization in at-risk patients (i.e., heavy smokers, immunocompromised patients). This rare localization is often pauci-symptomatic and thus can easily escape diagnosis. A case of isolated leishmaniasis limited to the left vocal cord in an immunocompetent Italian male without significant risk factors, randomly discovered upon histological examination, is described herein. We enquire how many patients affected by non-specific symptoms such as dysphonia and live in countries where Leishmania infantum infection is reported, could be truly affected by Leishmania spp infection.

REFERENCES