CASE STUDY

Laryngeal Leishmaniasis. A Case Report

Un caso de leishmaniasis laringea

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Clinical Case

We present the case of a 42-year-old male patient, smoker, diagnosed and treated for hepatosplenic leishmaniasis in 2008, without any travel to areas of risk.

He came for consultation 3 years later because of severe dysphonia (GRABS 3–4) with a 2-month history of dysphagia–odynophagia, previously unstudied.

Nasofibroscopy results were that the nostrils, oral cavity and pharynx are without lesions. Vocal cords were motile, of a greyish colour, with free rough border and mucous wave annulled under stroboscopy. There was diffuse oedema in the epiglottis and arytenoids (Fig. 1). Cervical palpation did not reveal anything of significance.

Serology was negative against VIH, VHC, and VHS 1–2 y lúes-leukocytosis lacking neutrophilia with PCR of 55 mg/l was found; ANA and ANCA were negative; Mantoux was negative; cultures of sputum and nasal and pharyngeal exudates were negative for Mycobacterium spp.

As the patient had not improved following a month of corticoid therapy, biopsies were taken using direct laryngoscopy, with histopathological report of Leishmania spp. amastigotes (Fig. 2). Leishmania infantum (L. infantum) was identified by DNA quantification of kinetoplast through PCR study on biopsies in culture.

Treatment with N-methylglucamine antimony parenterally was carried out for 15 days intrahospital, followed by every 15 days for 3 months.

At present the patient reports significant clinical improvement. Vocal folds appear with partially free borders and are functional, with moderate transmission of mucosa wave (GRABS 8–9).

Discussion

Leishmania is a flagellated protozoan, a blood and tissue parasite in canines and rodents, whose intermediate hosts are mosquitoes of the Phlebotomus or Lutzomya genus. It takes on various forms in its biological cycle: amastigote in the host, with nucleus and kinetoplast, from 2 to 5 μm of intracellular length; and promastigote in the digestive tube of the vector, then being longer and flagellate. It is transmitted to humans by the bite of an infected mosquito.

Dermotropical forms produce "Oriental button", Leishmania tropica (L. tropica), and other cutaneous-mucosal affectations (Leishmania braziliensis, infantum, amazonensis and mexicana). Laryngitis is especially rare.

Cutaneous-mucosal leishmaniasis is endemic in Asia, Africa, the Mediterranean and South America. In Brazil some 20 cases of mucosal forms are identified annually, compared with 500 cutaneous forms. With an incidence of 400 000 cases/year and world-wide prevalence of

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Laryngeal Leishmaniasis

12 million,\textsuperscript{1} the current mobility of humans, the variability of the incubation period and immunodeficiencies have propagated the zoonosis.

While cutaneous leishmaniasis shows a wide lesion range—ulcers with elevated borders or nodular forms that are more polymorphic and disseminated, and with greater nasal involvement—the mucosal forms offer a characteristic granulated surface image.

It is accepted that the mucosal affection can be through lymphatic and blood means or by direct cutaneous contact. Inhaling the parasite from the nostrils makes it possible to inoculate it in the upper airways.

Two leading Brazilian journals acknowledge that \textit{L. braziliensis} principally affects the nasal mucosa, with pharyngeal–laryngeal being the rarest form.\textsuperscript{1,2} In Europe, the condition varies according to species, with laryngeal involvement from \textit{L. infantum} being involved in between 35\% and 48\% of the cases.\textsuperscript{3,4} This lesion is similar to other mucosa; fibrinous membranes over swollen areas with transformation to wart-like granulations in epiglottis, vocal folds or ventricles. If it is a case secondary to cutaneous affection, the mucosal lesion develops over a mean time period of 264 days.\textsuperscript{1}

If there is clear endemic origin, immunodeficiency, prior visceral affection or concomitance of lesions in the nose and throat, this condition should be considered. The serology is very sensitive when the disease is suspected, while the intracutaneous reaction is not very useful due to the absence of cell response.

The granulations require biopsy to make a differential diagnosis with neoplasms and chronic granulomatous diseases. Once tuberculosis, leprosy, AIDS and syphilis are ruled out, microbiology and histopathology of the specimens should also assess possible affection by \textit{Paracoccidioides}, \textit{ Blastomycetes}, \textit{Actinomyces} and \textit{Histoplasma}. Biopsy offers as the most characteristic and most significant finding caseating granuloma, over the neovascularization and the macrophages with amastigote forms (easily recognisable with Giemsa staining, given that the kinetoplast becomes intensely red). Malignisation, amyloidosis, sarcoidosis and Wegner’s granulomatosis are also ruled out.\textsuperscript{5} If it is necessary to typify the species, the amastigote is sequenced using PCR.

Major dysphonia should be avoided by performing a biopsy on other affected areas if there are ones different from the vocal organ. The epiglottis produces odynophagia, and widespread lesions can provoke dyspnea.\textsuperscript{1,5}

Treatment is based on pentavalent antimonial (antimony gluconate and parenteral sodium or N-methylglucamine antimony) or intramuscular pentamidin, more effective and toxic. These agents are maintained from 3 days to 3 months, according to the severity of the symptoms.\textsuperscript{5} Intravenous administration ensures high concentration of antimony in blood, but it can be cancelled before or after this period based on patient response to therapy or degree of immunosuppression. Amphotericin B, more toxic, and imidazoles are reserved for resistant cases. Corticoid therapy can mask the disease for years. Topical application of agents and surgical excision of the lesions are valid alternatives in focalised lesions, including glottic ones, under direct laryngoscopy.\textsuperscript{1,2,6}

The sequelae of laryngeal leishmaniasis are secondary to fibrosis of aerodigestive pathways, with dysphonia from cancellation of Reinke’s space, subglottic stenosis and the possibility of permanent tracheotomy.\textsuperscript{5}

\textbf{Conflict of Interests}

The authors have no conflicts of interest to declare.
References