Case Report

Otolaryngologic Presentation of Mucosal Leishmaniasis

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Received 1 January 2013; Accepted 29 January 2013

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Abstract

Mucosal leishmaniasis (ML) is an uncommon complication of a disease seen primarily in the developing world. A 31-year-old previously healthy man presented with six months of nasal congestion, throat pain, hoarseness, and intermittent dysphagia and odynophagia. The leishmania culture was positive, and PCR testing was consistent with Leishmania braziliensis infection. He was treated with liposomal amphotericin B therapy for three weeks. He resolved symptomatically after therapy. This case highlights the necessity of obtaining accurate travel and social history and the often delayed presentation of mucosal leishmaniasis.

Keywords

leishmaniasis; mucosal leishmaniasis; dysphagia; odynophagia; omega epiglottis; nasopharyngeal swelling; oropharyngeal swelling; leishmania

1. Introduction

Mucosal leishmaniasis (ML) is an uncommon complication of a disease seen primarily in the developing world. We report a case of advanced ML in a patient in the United States. Our case highlights the importance of a complete travel history and appropriate consultation in patients with diffuse mucosal inflammation.

2. Case presentation

A 31-year-old previously healthy man presented to the Otolaryngology—Head and Neck Surgery clinic with six months of nasal congestion, throat pain, hoarseness and intermittent dysphagia and odynophagia. He was a Brazilian immigrant and worked in landscaping.

On examination, he had diffuse inflammation and an irregular appearance of the mucosa in his nasal cavity, oropharynx, and supraglottic larynx (Figure 1). Flexible fiberoptic nasolaryngoscopy demonstrated a thickened omega-shaped epiglottis (Figure 2) and thickening and irregularity of the supraglottic mucosa, including the aryepiglottic folds, arytenoids, and false vocal folds. His physical exam was otherwise benign; he had no rashes or skin lesions.

He was initially treated with ciprofloxacin for suspected gonococcal infection. Diagnostic testing included a non-reactive PPD, and negative AFB, fungal, PAS, and GMS culture. Complete blood count, serum electrolytes, ESR, CRP, ANA, c-ANCA, and p-ANCA were normal. An oropharyngeal mucosal biopsy showed squamous mucosa with acute and chronic necrotizing granulomatous inflammation and no malignancy. Consultation from the Infectious Disease service was obtained. The differential diagnoses included leishmaniasis, paracoccidiomycosis, and malignancy. A second biopsy of nasal and oropharyngeal tissue was obtained, and specimens were sent to the Center for Disease Control for leishmania culture and repeat fungal and bacterial cultures. The leishmania culture was positive, and PCR testing was consistent with Leishmania braziliensis infection.

He was admitted for initiation of therapy. Because of concerns about increased swelling and airway compromise with treatment, he underwent elective tracheostomy prior to beginning liposomal amphotericin B therapy. He responded well, with his course complicated by an episode of hypokalemia.

Figure 1: Oral cavity showing irregular thickening.
3. Discussion

Leishmaniasis is a parasitic protozoal infection transmitted by sand flies and is estimated to affect 10–20 million people worldwide, with approximately 2 million new infections per year [5]. The burden falls primarily on developing countries, but the disease is found on all inhabited continents [5]. The reservoir includes humans, and both wild and domesticated animals. Most cases in the Western Hemisphere are found in South America, and it is not considered endemic to the United States or Canada [2,5]. Incidence among travelers from developed countries is increasing [3]. There are two primary classifications—cutaneous and visceral, where the parasite resides in dermis and macrophages, respectively [2]. The cutaneous form can also progress to mucosal infection, occasionally without any cutaneous manifestations [5]. Approximately 75% of new cases present with mucocutaneous lesions [5].

After the initial sand fly bite, the most common course involves a papule that develops into a painless ulcerated nodule over weeks to months [5]. Satellite lesions may occur. This generally heals within a year, depending on the species. ML, seen in this patient, develops in approximately 3% of cases and is caused almost exclusively by American continent species of *Leishmania braziliensis, Leishmania amazonensis, Leishmania guyanensis,* and *Leishmania panamensis* [5]. It is associated with diffuse cutaneous leishmaniasis, a manifestation seen in individuals with poor cell-mediated immune response to the pathogen [5].

ML is characterized by spread to the upper respiratory tract mucosa. It often occurs 1–5 years after the initial infection but can appear concurrently or after decades [5]. Initial manifestations involve the mouth and nose, followed by the pharynx and larynx in advanced disease. Typical symptoms begin with rhinorrhea and epistaxis followed by septal perforation/nasal cartilage collapse, dysphagia, odynophagia, and dysphonia [2]. Respiratory compromise is the predominant cause of fatality (rare) in ML [1].

ML will not resolve spontaneously and requires treatment, generally with amphotericin B in the United States, and sodium stibogluconate or meglumine antimoniate elsewhere [5]. Other medications and combinations have been used [2], and infectious disease consultation is advised. Tracheostomy may be considered for patients at risk of airway compromise.

4. Conclusion

Previous reports in dental literature have highlighted oral complications [1]. One past report highlighted the possibility of isolated dysphonia from visceral leishmaniasis [4]. Leishmania is well known in countries where it is common. However, it is much less commonly reported in American literature. It is important for otolaryngologists and others who may be consulted for this condition to be aware of its presentation. It should be considered in the differential diagnosis of diffuse mucosal inflammation, particularly in immigrants from South America.

References


