CASE REPORT

LARYNGEAL LEISHMANIASIS IN A 46-YEAR-OLD ETHIOPIAN PATIENT

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ABSTRACT

Leishmaniasis is a vector-borne zoonotic disease caused by intracellular protozoa of the genus Leishmania. Leishmania species produce widely varying clinical syndromes ranging from self-healing cutaneous ulcers to fatal visceral disease. Clinical presentations of Leishmania infection include visceral (most common form), cutaneous, mucocutaneous, mucosal and post-kala-azar dermal leishmaniasis. Mucosal form of leishmaniasis mostly involves oral and nasal mucosa. Rarely, laryngeal and pharyngeal mucosa may also be involved.

In this presentation, we report a 46-year-old male patient, from Gondar, Northern Ethiopia. He developed progressive hoarseness, dysphagia, loss of appetite and weight loss over eight months duration. He had swelling of the upper lip and splenomegaly. Cervical CT showed laryngeal mass and biopsy from the mass revealed Donovan bodies. The patient was treated with standard anti-leishmania regimen, and showed significant clinical improvement.

Keywords: Leishmaniasis, Splenomegaly, Dysphonia, Kala-azar

INTRODUCTION

Leishmaniases are parasitic diseases caused by protozoan flagellates of the genus leishmania and transmitted by the vector phlebotomine sand fly (1). They occur in 98 countries-most of them developing-in tropical and temperate regions (2). Traditionally, the disease is classified into three clinical presentations: visceral (VL), cutaneous (CL) and mucocutaneous (MCL) (3). In Ethiopia, both the cutaneous and visceral forms of Leishmaniasis are endemic (8). MCL is not commonly reported in Ethiopia (5).

MCL is usually secondary to hematogenous spread after months or years of skin infection and can manifest as an infiltrative lesion, ulcerated or vegetating lesion in the nose (most common site), pharynx, larynx and mouth with or without lymphadenopathy (6). Typically, MCL presents as nasal stuffiness and bleeding followed by destruction of nasal cartilage, perforation of the nasal septum, and collapse of the nasal bridge. Subsequent involvement of the pharynx and larynx leads to difficulty in swallowing and phonation (2).

Laryngeal extension follows the rhino-buccalpharyngeal localization of the parasite. The lesion initially manifests as dysphonia and metallic cough (1). Even though isolated laryngeal leishmaniasis is an uncommon feature of this zoonotic disease, the larynx could be affected if the upper respiratory structures such as nasal cavity are involved first, due to spread of the infection to the larynx and laryngeal mucosa. This is typical of Leishmania braziliensis infection, which is common in South America (7). Here, we present a case of laryngeal leishmaniasis in a 46-year-old immunocompetent farmer from Humera, an endemic area for visceral leishmaniasis (8).

CASE REPORT

A 46-year-old male Ethiopian from Humera, Gondar, initially noticed a small swelling in the left nostril, which was then followed by hoarseness of voice, progressive difficulty in swallowing of solid foods, anorexia and weight loss over eight months duration. He had dragging sensation on his left upper abdomen. He also had a dry cough for 6 months.

Physical examination revealed an erythematous swelling on the upper lip. There was an enlarged spleen measuring 8cm below the left costal margin. Examination of the other systems revealed no abnormalities.

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On investigation, complete blood count, liver and kidney function tests were normal. Serology for HIV was negative. CT-scan of the neck showed a soft tissue mass involving the true vocal cords bilaterally with irregular outline, with involvement of the aryepiglottic folds, the free margin of the epiglottis on the right, the vallecula and pyriform sinuses (Figure 1).

Fiberoptic laryngoscopy was done and the findings revealed a swollen epiglottis with posterior laryngeal edema. With an impression of laryngeal carcinoma, the ENT team performed surgery. The intraoperative finding showed a polypoid mass occupying the vallecula and pyriform sinuses extending to the aryepiglottic folds with an anterior commissural hanging pale mass with papillomatous growth involving the right vocal cord. Biopsy was taken from the glottis lesion. Microscopic examination revealed squamous epithelium displaying low grade dysplasia and underlying intense lymphohistiocytosis and intracellular Donovan bodies. Splenic aspiration for visceral leishmaniasis was unrevealing.

After tissue diagnosis, the patient was treated with paromomycin for five days and sodium stilbogluconate for two months. With this treatment, the patient showed significant clinical improvement evidenced by improvement of dysphagia and dysphonia. Repeat neck CT and laryngoscopic examination were not done due to financial limitations.

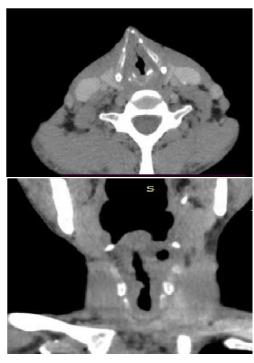


Figure 1. Non contrast CT-scan of the Neck, showing soft tissue mass involving the bilateral true vocal cords with irregular outline, the ariepiglotic folds and the free margin of the epiglottis on the right.

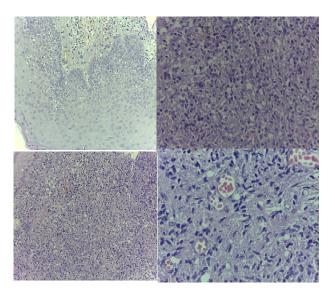


Figure 2. Microscopic-histologic examination showing squamous epithelium and underlying intense lymphohistiocytosis and intracellular Donovan bodies

DISCUSSION

In Ethiopia, VL has occurred as epidemics in the northwestern part of the country, bordering Sudan, such as Humera and Metema. Due to high mortality, occurrence of epidemics, and high incidence of the disease, leishmaniasis has become one of the leading health problems in Ethiopia (7). Sporadic cases of CL have been diagnosed from many localities in the Northern, Central, and Southern high lands of Ethiopia (7). In Ethiopia, cases of mucocutaneous leishmaniasis, including laryngeal leishmaniasis are rare. To authors' knowledge this is the first case report from Ethiopia on biopsy proven laryngeal leishmaniasis.

To date, only few cases have been reported on laryngeal leishmaniasis. Kamran and his colleague reported a case of mucosal leishmaniasis on a vocal cord without systemic involvement in a patient with hoarseness of voice and difficulty in breathing (8). Kumar and his colleague also reported a patient with a subglottic mass with a final diagnosis of subglottic leishmaniasis in which the patient presented with hoarseness, shortness of breath and cough for 3 months duration (9).

Our patient presented typically with progressive hoarseness, difficulty swallowing and constitutional symptoms over 8 months, with imaging evidence of a mass on the vocal cords. Since Ethiopia is one of the sub-Saharan countries where tuberculosis is endemic, it is important to rule out laryngeal tuberculosis, which can present with the same clinical feature. The

normal chest x-ray, normal ESR and complete blood count and above all response to anti-leishmanial drugs make tuberculosis less likely in this patient.

Conclusion: Laryngeal leishmaniasis should be considered in the differential diagnosis in patients presenting with progressive hoarseness and difficulty of swallowing, more so in patients from geographical areas where leishmania epidemics have been reported.

Abbreviations: VL-Visceral Leishmaniasis, MCL – Mucocutanous leishmaniasis, ENT – Ear, nose and throat.

ESR – Erythrocyte sedimentation rate.

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