

Laryngeal Leishmaniasis- A Rare Clinical Entity

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Abstract Isolated laryngeal leishmaniasis is a rare condition and we report a 46 years old patient who presented with progressive, non fluctuating hoarseness of voice for 8 months along with foreign body sensation of throat and difficulty in swallowing. Indirect laryngoscopy followed by flexible nasoendoscopy revealed grossly edematous epiglottis, right arytenoid, right aryepiglottic fold and medial wall of the right pyriform sinus. Direct laryngoscope was performed under general anesthesia and biopsy was taken. The histopathology report section revealed numerous extracellular L.D. bodies in an inflammatory milieu comprising of lymphocytes, neutrophils and macrophages set in an edematous subepithelial stroma. He has no other systemic manifestation. He had no HIV infection or immunocompromised condition nor took immunosuppressive drugs. He responded well with liposomal Amphotericin B.

Keywords: *leishmania donovani*, LD Bodies, larynx

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1. Introduction

Leishmaniasis is a worldwide infection with a broad spectrum of clinic pathological features. [1] Leishmaniasis is caused by a protozoan parasite of the Genus *Leishmania*. The parasite is transmitted by the bites of sand fly infected with the parasite. [2] Clinical leishmaniasis presents in the three main clinical forms-coetaneous (C), Mucocutaneous (MC) and visceral (VL- KALA-AZAR) with a broad range of symptoms, signs and degree of the severity. Endemic foci of leishmaniasis are found in all continents except Australia and Antarctica. [3,4] There are more than 20 species that are divided into old and new world types. These include the *L. donovani* complex with 2 species (*L. donovani*, *L. infantum* [also known as *L. chagasi* in the New World]); the *L. mexicana* complex with 3 main species (*L. mexicana*, *L. amazonensis*, and *L. venezuelensis*); *L. tropica*, *L. major*, *L. aethiopica* and the subgenus *Viannia* with 4 main species (*L. (V.) braziliensis*, *L. (V.) guyanensis*, *L. (V.) panamensis*, and *L. (V.) peruviana*). [5] Based on geography leishmaniasis is divided in to old world type and new world type. New old type is found in some parts of Asia, the Middle East, Africa (particularly in the tropical region and North Africa, with some cases elsewhere), and southern Europe. [6] Old world type is found in some parts of Mexico, Central America, and South America [6].

We report a case of primary laryngeal leishmaniasis presented with hoarseness and confirmed with the histopathological examination of the tissue taken from direct laryngoscopic biopsy from right arytenoids and aryepiglottic fold.

2. Case

A 46 years old patient from endemic region of leishmaniasis presented with a hoarseness of voice for 8 months which was insidious onset, gradually progressive, non fluctuating and painless. It was associated with foreign body sensation in the throat and difficulty in swallowing.

There was no significant past medical and surgical history.

He consumes alcohol and was a smoker (15 pack years).

On general examination no abnormality was detected and on indirect laryngoscopy followed by flexible nasoendoscopy revealed grossly edematous epiglottis, right arytenoid, right aryepiglottic fold and medial wall of the right pyriform sinus.



Figure 1. Edematous Epiglottis on flexible naso-endoscopy



Figure 2. swollen right arytenoid and right aryepiglottic fold on flexible naso-endoscopy.

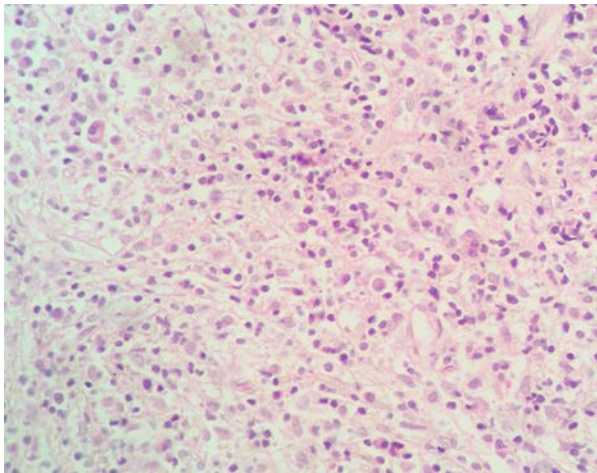


Figure 3. Histopathology showing LD bodies in 400 X

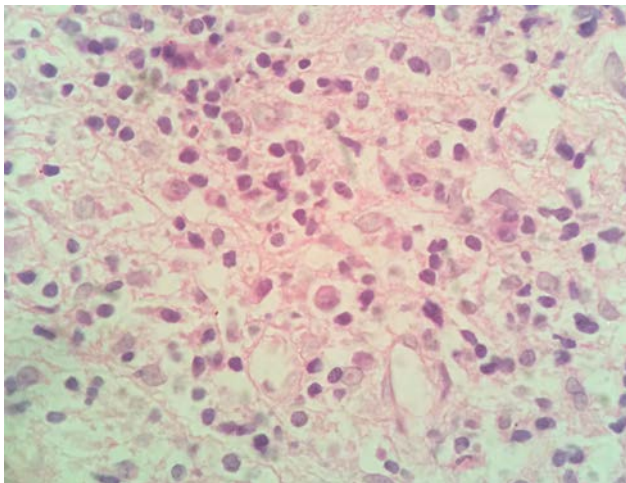


Figure 4. Histopathology showing LD bodies in oil immersion microscopy

Direct laryngoscopic biopsy was done under general anesthesia and the specimen sent for histopathology revealed numerous extracellular L.D. bodies in an inflammatory milieu comprising of lymphocytes, neutrophils and macrophages set in an edematous subepithelial stroma. Identification of species was not possible as culture of leishmania was not available in our set up. He had no other systemic manifestation, HIV infection or immune-compromised condition nor took immunosuppressive drugs. He was treated with liposomal

AmphotericinB (3 mg/kg from day1 to 5, and further dose on day 14 and 21). [7] We used Evocin of Ranbaxy Laboratories Limited and it was available at hospital pharmacy. After treatment the patient was reevaluated at 6 weeks and there was marked improvement in symptoms with no hoarseness and flexible nasendoscopy showed minimal edema of right arytenoid.

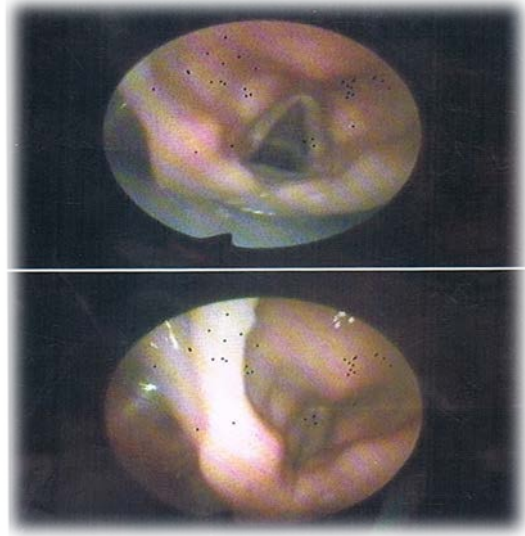


Figure 5. Post treatment picture of flexible endoscopy showing normal larynx

3. Discussion

Leishmania donovani was discovered in 1900 by William Fleishman (Scottish army doctor) and Charles Donovan (professor of physiology at Madras University, India) separately in the spleen of the patient with kala-azar. [8] The vector was indentified after long journey in 1921 with a experimental proof of transmission to human by the bite of the sandfly of genus *Phlebotomas* by Edouard and Etienne [9,10].

Leishmania is an obligate intracellular parasite infecting the macrophages of the host with a variety of clinical manifestation of CL, MC and VL. In vertebrate they are found in the amastigote form *Leishmania Donovan* (LD) bodies. They replicate into the cells of the reticulo-endothelial system. The parasite enters into the gut of the sandfly and develops into promastigote and migrates to salivary glands and is inoculated to the innocent new host. [11] The mucosal disease is usually associated with the coetaneous and visceral disease. However HIV and other immunosuppressive condition like malnutrition, steroid therapy, chemotherapy, chronic alcohol and smoking are the risk factors for the disease [4,8,11].

Mucosal leishmaniasis is a rare disease and few cases reported involving oral mucosa and mucosa of the upper respiratory tract. [13] Nasal septum was found to be the most common site and present with epistaxis, with destruction of the pharynx and larynx. [14] Clinically evident leishmanial infection of the naso-oropharyngeal mucosa is the potentially disfiguring metastatic complication the coetaneous leishmaniasis [15].

Treatment with pentavalent antimonial compound is moderately effective for the mucosal disease and

Amphotericin B can be considered the first line treatment in mucosal disease. Patient with the respiratory compromise can be co-administered with the glucocorticosteroids. [15]

Very few cases have been reported in laryngeal leishmaniasis. Trevor et al., 2012 reported the vocal cord leishmaniasis in 84 years old man with hoarseness of voice. [3] Kamran et al., 2005 reported the mucosal leishmaniasis on a vocal cord without systemic involvement in patient with hoarseness and difficulty in breathing. [10] Kumar et al., 2009 reported a 70 years old male with suspicious of the subglottic mass with a final diagnosis of subglottic leishmaniasis in patient with hoarseness of the voice, dyspnoea and cough for 3 months duration. [8] Our patient also presented with similar complain of hoarseness with discomfort of throat and difficulty in swallowing.

4. Conclusion

Laryngeal leishmaniasis should be considered one of the differential diagnoses of laryngeal mass besides carcinoma and tuberculosis in endemic zone of leishmaniasis.

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