EDITORIAL

Hypopharyngeal Leishmaniasis: A case Report and Literature Review.

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Abstract:

Background: Leishmaniasis is classified into three clinical patterns: visceral, cutaneous and mucocutaneous.The latter are sporadically reported in Sudan and may not accompany the visceral type. Laryngeal symptoms including dysphagia may mimic oesophageal malignancy. Early detection and proper diagnosis are of great help in the cure and prognosis of the disease.

Case report: We reported a case of primary isolated (the nasal mucosa was not involved) Hypophangeal Leishmaniasis in an immunocompetent Sudanese patient presenting with dysphonia, odynophagia and progressive dysphagia.

Conclusion: Leishmaniasis should be suspected in all patients presenting with laryngeal symptoms in patients coming from endemic areas.

Keywords: Hypopharyngeal lesion, leishmania, Leishmaniasis, Sudan.

Introduction:

Hypopharngeal leishmaniasis is rarely encountered as an isolated lesion. However, nasopharyngeal, retropharyngeal and oral lesions of Leishmaniasis are found sporadically in Sudan and East Africa and have been described in India[1,2].They are found in areas where Kala-azar occurs and may, or more commonly may not be associated with the visceral disease. Mucosal leishmaniasis (ML) is an unusual immunological response in certain individuals to infection with L.infantum. The causative Leishmania is identical to that causing Kala-azar[3]. In Leishmaniasis, the larynx is more often involved, when mucosal lesions are firstly localized to the nose and /or oral cavity, they may descend in the upper respiratory tract and may involve the laryngeal mucosa[4].Here, we report a case of primary isolated laryngeal (no nose involvement)Leishmaniasis in an immunocompetent Sudanese patient.

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Case Report:

A 30 years old housewife,from Gedarif East Sudan, presented to our hospital with a 6-months history of a progressive dysphagia to solids and then to liquids. She equally reported a high-grade intermittent nocturnal fever with night sweating and she noticed some weight loss. She gave a history of dysphonia, but no nasal regurgitation or dysarthria.Her musculoskeletal system was free from arthritis or skin rash. She had no similar condition among her family members. The patient was not on any medication, not smoker, and not used to consume alcohol. During the examination, the patient was ill, wasted, slightly pale, but not jaundiced or cyanosed. Her chest, cardiovascular system abdomen, and central nervous system (CNS) were all unremarkable.

Investigations:

Laboratory studies revealed a normal hematocrit, hemoglobin, white blood cell count, differential, platelet and red cell morphology. Erythrocyte sedimentation rate was 77mm/h. Liver and renal function tests were normal. Serological tests were negative for human immunodeficiency virus (HIV). The bone marrow aspiration/examination was negative for L.donovani bodies (LD bodies). Abdominal ultrasound showed no organomegaly. Chest X-Ray showed no features of tuberculosis.

Telelaringoscopy was done by an ENT surgeon and revealed a mass in the right piriform fossa. A biopsy was taken and showed microscopically an extensively ulcerated hyperkeratotic stratified squamous epithelium with severe reparative atypia, necrosis, bacterial colonies, dense mixed inflammatory infiltrate and the presence of Lieshman Donovani bodies (L.D bodies). No evidence of malignancy was seen in the adequate biopsy material submitted. Given this histopathology report, laryngeal leishmaniasis was suggested in spite the non-involvement of the nose and oral cavity. Seven days later a flexible esophagoscopy was done that revealed an ulcerating mass in the right piriform fossa, at 10 cm from the incisor tooth (Fig 1).Biopsy/histopathology examination report confirmed the initial biopsy diagnosis. Unfortunately we failed to include the histopathology picture because it was lost.

Treatment:

Based on the above findings, a baseline Electrocardiogram (ECG) was obtained, and intravenous sodium stibogluconate was started (ALBERT DAVID, INDIA) at a dose of 20mg/kg daily for 30 days according to the World Health Organization recommendations[5]. The fever subsided with substantial improvement regarding hoarseness of voice and dysphagia.

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Figure(1): Flexible esophagoscopy showing an ulcerated hypopharyngeal mass.

Discussion:

The presented case was a 30-year old immunocompetent woman with a rather atypical presentation of hypopharyngeal leishmaniasis in Sudan[6].She reported no history of visceral leishmaniasis (VL) before. Hypopharyngeal leishmaniasis could follow an episode of VL or can be a primary disease as seen in our case [7].The commonly presented cases in Sudan were isolated mucosal forms not usually accompanied by mucocutaneous lesion [6].

Many have assumed that both the visceral and the mucosal forms are caused by the same parasite[8]. However, Abdelhamid *et al*. reported that mucocutanous leishmaniasis (ML) in Sudan is due to a different subspecies of the L.donovani complex that causes VL[9].

The laryngeal lesions usually show swollen vocal cords with an irregular surface and white patches [10].In our case the involvement was intense, the patient had a huge mass filling the right piriform fossa. The growth significantly affected eating and nutrition. In many cases and because of delay of medical care such growths might endanger the patient life through airway obstruction. There was no evidence of other granulomatous disorders like leprosy or tuberculosis. The absence of the involvement of the nose (as the initial presentation of mucosal leishmaniasis) is extremely rare.

Our patient was from Eastern Sudan (Gedaref) which is a known endemic area for both VL and HIV infection. The Sudan AIDS Program(SNAP) reported that the prevalence of HIV/AIDS is 1.6% among the general population [11]. An association between ML and HIV has been described [12]. Leishmaniasis can develop at any stage of HIV infection with clinical manifestations coinciding with the maximum immune depression. The current case was tested negative for anti-HIV antibodies.

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No standardized protocols are available for the treatment of laryngeal Leishmaniasis because of the small number reported worldwide, both liposomal amphotericin B and pentavalent antimonials seem to be effective, although the dose and duration are different[7,10].

In conclusion: Although rare, Leishmaniasis should be suspected in all patients presenting with laryngeal symptoms especially in those coming from endemic areas.

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