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Old World Mucosal Leishmaniasis Treated with a Single Dose of Amphotericin: A Case Report

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ABSTRACT

Mucosal leishmaniasis, an uncommon disease in the old world, is recognized as an occasional complication of donovani related visceral leishmaniasis. It usually affects the spleen, liver, skin, mucosa of the respiratory tract and, less commonly, the oral cavity. Our case was a young adult with non-healing lesions in the oropharyngeal region who was diagnosed as mucosal leishmaniasis during evaluation for suspected mucosal carcinoma. The diagnosis was made with the rK39 strip-test after multiple futile test results. A single dose of liposomal amphotericin B cured the disease.

Keywords
Leishmania donovani; leishmaniasis; mucosal

INTRODUCTION

Leishmaniasis has three major forms: cutaneous leishmaniasis (CL), visceral leishmaniasis (VL, aka kala-azar) and mucocutaneous leishmaniasis (MCL).[1] Mucosal leishmaniasis (ML) primarily affects the mucosa of the upper respiratory tract and the oral cavity.[2] Although ML is primarily linked to *Leishmania braziliensis* (South America) and *L. infantum* (Mediterranean basin), cases have been reported with with *L. donovani* in India, Sudan, and Sri Lanka.[2,3] A minority of patients with Post kala-azar dermal leishmaniasis (PKDL) may have mild mucosal involvement.[4] In the old world variant, mucosal disease can be preceded or accompanied by CL or VL.

Nepal is one of the five endemic countries for VL globally and most of the elimination efforts are being focused on VL-endemic areas of Nepal.[5] Isolated mucosal leishmaniasis is rare in Nepal and our case is of isolated mucosal leishmaniasis successfully treated with single dose of liposomal Amphotericin B.

CASE PRESENTATION

A 33 years old young male adult from Makwanpur presented with fever and difficulty swallowing for seven days. The patient had a past medical history of non-healing lesions in the oropharynx for four years. There was also a history of discharge from the nasal cavity, increased salivation, and difficulty swallowing solid and liquid foods for the past seven days.

Four years before the presentation, the patient had developed growth posterior to his right 3rd molar. The histopathology done from the retromolar growth had shown stratified squamous epithelium with numerous plasma cells and few lymphocytes. Serum protein electrophoresis was normal. He had also been treated for pulmonary tuberculosis for six months. The oropharyngeal lesion was still present.

On examination, an ulceroproliferative lesion was seen over the soft palate and oropharynx without cervical lymphadenopathy. Nasopharyngolaryngoscopy revealed an ulcerative lesion covered with slough involving uvula, nasopharynx, lateral pharyngeal wall and hypopharynx. There was no organomegaly, skin discoloration or scars on physical examination. Hematological, biochemistry and serological findings were normal. Throat swab revealed growth of *Klebsiella pneumoniae* but treatment did not improve the symptoms completely.

Biopsy from the soft palate showed dermis infiltrated with mixed inflammatory cells (plasma cells, lymphocytes, histiocytes) with plasma cell predominance. (Figures 1 and 2) and immunohistochemistry favored inflammatory pathology with positive immunostaining for CD138, Kappa light chain, lambda light chain, CD3, and CD20.

Bone marrow aspirate was normal. Ultrasonography of the abdomen and pelvis showed liver of 15 cm and spleen of 12.2 cm. Chest X-Ray showed fibrotic changes in the right upper zone, likely the sequelae of past PTB. Finally, the rK39 strip showed a positive result and the diagnosis of mucosal Leishmaniasis was made (Figure 3). The patient improved after receiving a single dose of intravenous liposomal amphotericin B infusion (Figure 4). The last follow-up was done via telephone interview two years later and there was no relapse.

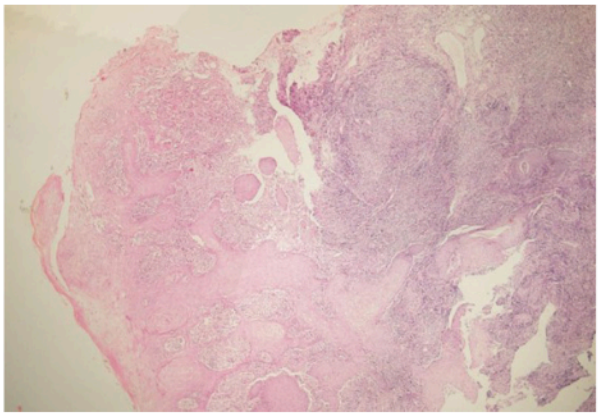

Figure 1. *Histopathological findings of soft palate – stratified squamous epithelium with underlying inflammatory infiltrates*

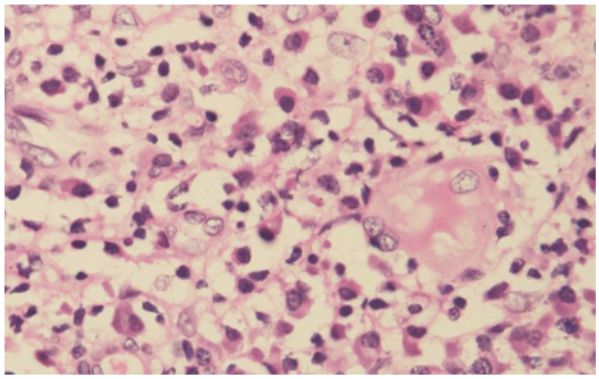

Figure 2. *Magnified view showing inflammatory cells composed of plasma cells, lymphocytes and histiocytes*

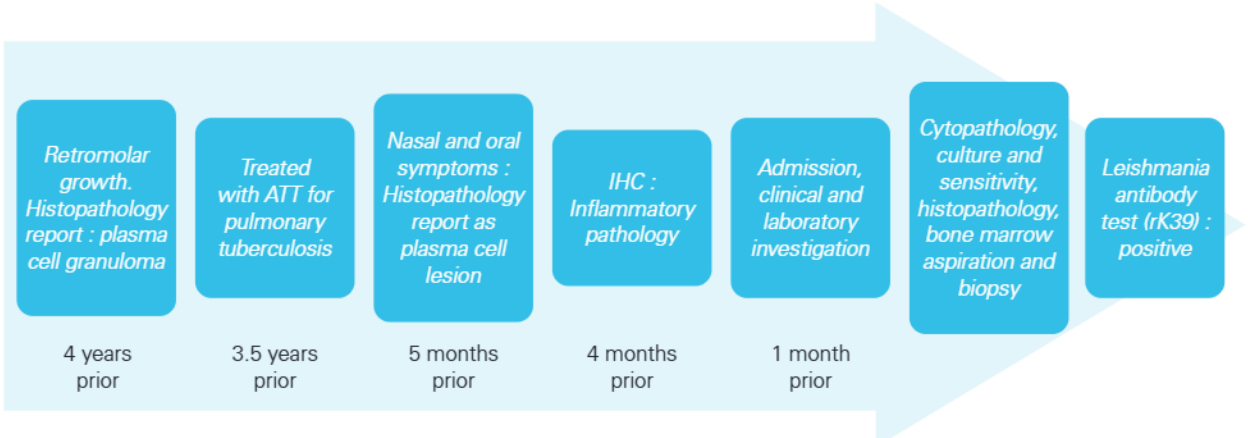

Figure 3. *Timeline of sequence of events in patient's history and investigation*

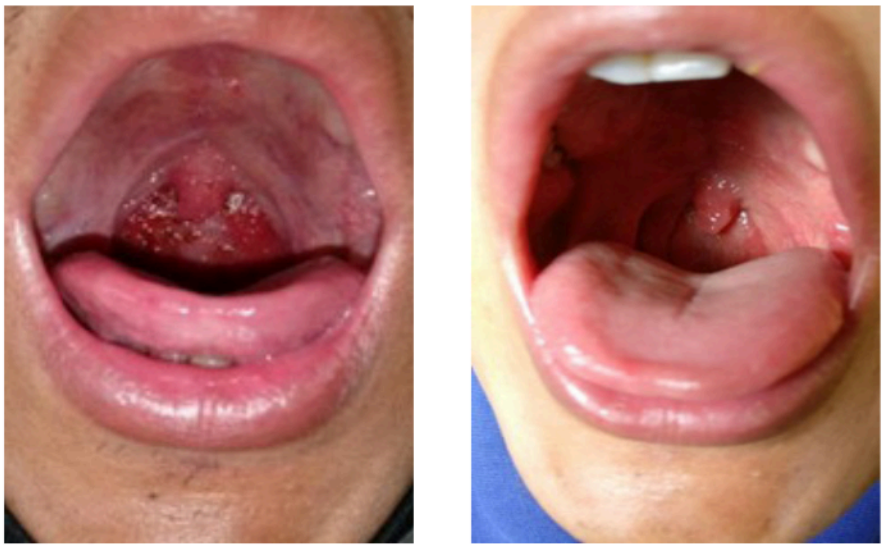

Figure 4. *Changes in mucosal lesion before and after the treatment*

DISCUSSION

In the Old World, mucosal leishmaniasis is considered a rare complication of VL (in Africa) or post-Kala-azar CL (in India) and isolated ML is even rarer.[6] ML is frequently misunderstood and underdiagnosed by physicians and scientists, especially outside of the South America, due to its diverse appearance.[2]

The mechanism of mucosal involvement in *L. donovani* infection is unclear and could be the site of parasite inoculation or secondary localization either by direct extension of contiguous skin lesions or via lymphatic/hematogenous spread from a distant site.[4] *Leishmania donovani*, as well as *Leishmania infantum*, have been reported to cause localized mucosal disease in the absence of concomitant visceral or cutaneous leishmaniasis.The histological diagnosis of ML is based on the demonstration of leishmania amastigotes in mucosal samples stained with Giemsa or hematoxylin-eosin stains.[2] Our patient also went through several histopathological tests but without getting a definitive diagnosis. This aspect emphasizes the importance of complementing the evaluation with a further diagnostic procedures such as ELISA or PCR in all uncertain cases.[5,7]

ML caused by *L. donovani* is a rare but increasingly reported entity from the endemic regions, especially among immuno-compromised subjects.[2] In our patient, throat swap was positive for *Klebsiella pneumoniae* suggesting secondary bacterial infection. The presence of antibodies against Leishmania, mild hepatosplenomegaly, and resident of *L. donovani* endemic zone suggest that the patient might have acquired subclinical visceral leishmaniasis in the past, which after many years of latency, got activated and manifested as a mucosal lesion.

In Nepal, the treatment of choice for VL is a single-dose of liposomal amphotericin B (L-AmB) but there no clear guideline in the treatment of ML. Therapeutic options for the treatment of ML include parenteral pentavalent antimony drugs, amphotericin B including liposomal amphotericin, and miltefosine.[8] However, our patient was cured with a single dose of L-AmB (10 mg/Kg) with complete resolution of the lesion in four weeks.

Although our patient was a resident of endemic zone for *L. donovani* and antibodies to the rK39 antigen were positive, we could not demonstrate the presence of the species (*Leishmania donovani* complex) within the lesion.

CONCLUSION

Although ML caused by *L donovani* is rare, health care providers should be aware and have a high index of suspicion in patients with persistent oral ulcers that do not respond to treatment. Early diagnosis and treatment with anti-leishmanial can prevent irreversible damage to the mucosal tissues. Considering the likelihood of relapses, the patient should be followed-up for some years.

CONSENT

Written informed consent was taken from the patient for the case report publication.

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CONFLICT OF INTEREST

The author(s) declare that they do not have any conflicts of interest with respect to the research,

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AUTHORS CONTRIBUTIONS

ST and MT did literature review. ST, MT, PP and AS collected the patient data. MT prepared the first draft. ST, PT and PP reviewed and revised the manuscript. All authors contributed to the final version of the manuscript.

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