Case Report

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Mucosal Leishmaniasis: a Rare Infection from Western Nepal

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Abstract

Leishmaniasis is a neglected tropical disease infecting world's poorest population in over 90 countries throughout Asia, Africa, the Middle East, and Central and South Africa. An underestimated 700,000 to one million new cases occur annually. Leishmaniasis refers to a spectrum of diseases caused by a parasite, Leishmania, transmitted by bite of infected sandflies. Of the three major syndromes (cutaneous, mucosal and visceral) mucosal leishmaniasis is the least common. Despite being an endemic country for cutaneous and visceral forms, there are rare reports of mucosal form published in the literature from Nepal till date. Here we present a case of mucosal leishmaniasis presented masquerading malignancy from Dailekh District immediate proximal to Surkhet, one of the endemic districts of western Nepal.

Keywords: Leishmania; Leishmaniasis, Cutaneous; Leishmaniasis, Mucosal; Leishmaniasis, Visceral.

Declarations

Ethics approval and consent to participate: Not applicable

Consent for publication: Informed consent was obtained from the patient's parents for the publication.

Availability of data and materials: Data will be made available upon request.

Competing interest: None

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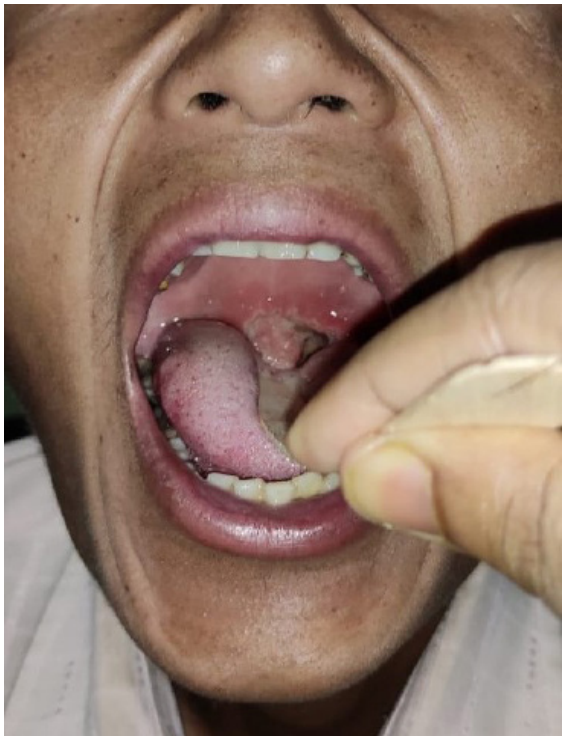
Authors contributions: PB: intellectual content, literature search, manuscript finalization. SCM: Provided clinical details along with photographs. MA: Review of literature, photography of the histology slides. RS: Review of literature, manuscript drafting. PGG: Critical review of the manuscript. All the authors have read and approved the final manuscript..

Leishmaniasis is a complex zoonotic disease caused by multiple subspecies of *Leishmania.* It is a vector borne obligate intracellular protozoan parasite, transmitted by bite of infected sandflies of *Phlebotomus* and *Lutzomyia* species. Diverse clinical manifestations range in each of three forms namely Cutaneous Leishmaniasis (CL), Mucosal/ mucocutaneous Leishmaniasis (ML/ MCL) and potentially fatal Visceral Leishmaniasis (VL), which leads to confusion even among the experts. ML indicates involvement of mucosa of upper respiratory tract and oral cavity. Typically, it manifests from days to years after CL, can either be accompanied or preceded by CL or VL [1].

1A



1B

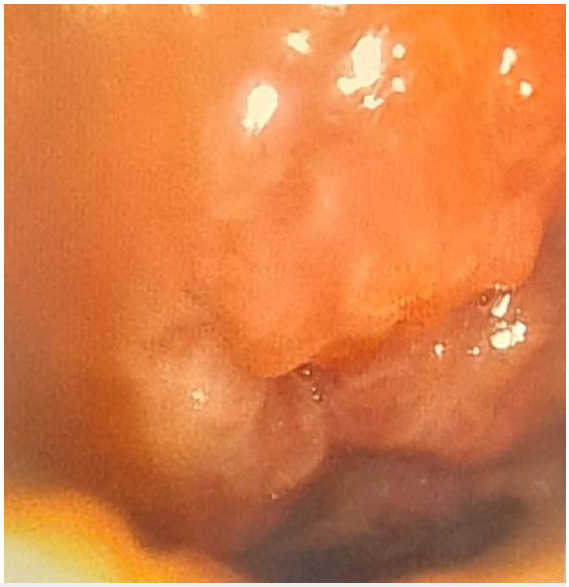


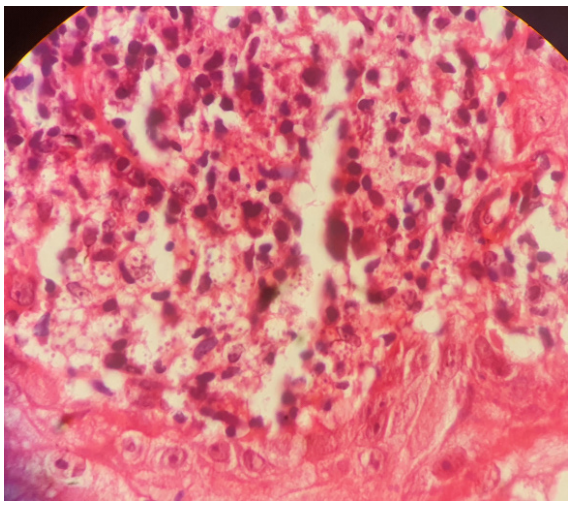
Figure 1: A. Ulceroproliferative growth in the posterior wall of oropharynx with swollen uvula. B. Nasal endoscopy showing swollen mass of whitish ulceroproliferative growth.

CASE

A 26 years old male, native from Athbiskot, Dailekh, Nepal presented to ENT department with throat pain, nasal obstruction accompanied by dysphagia for over 5 - 6 months. On nasoendoscopic examination multiple pale whitish masses were found in nasopharynx filled up to oropharynx, the largest one measuring approximately 3x4 cm in the oropharynx **(Fig. 1A and B).**

He had no significant history of any past illness. He was a farmer by occupation. On examination, bilateral cervical lymphadenopathy and a depigmented round and retracted scar on his left forearm of 5 years duration were revealed. His systemic examination was also unremarkable.

2A



2B

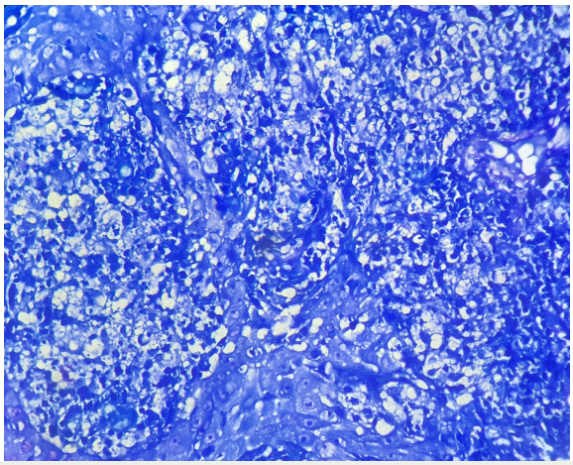


Figure 2: A. Mucosal biopsy, submucosa showing macrophages with numerous amastigotes of Leishmania species (HE stain, X1000). B. Giemsa staining demonstrating the same (X400).

Multiple punch biopsies from oropharynx were taken and sent for histopathology. Histopathological examination from oropharyngeal tissue revealed hyperplastic mucosa with areas of erosion. Submucosa showed diffuse infiltration of histiocytes, plasma cells and lymphocytes

forming ill-defined granulomas. Histiocytes- macrophages demonstrated occasional small round uniform intracellular organisms of Leishmania amastigote (L. D. Bodies) in their cytoplasm stained on Hematoxylin & Eosin and Giemsa **(Fig. 2A and B)**. Ziehl Neelsen and Periodic Acid-Schiff (PAS) stain were both negative. Hence, the diagnosis of ML was made.

Routine blood investigations including Complete Blood Count, Peripheral Blood Smear, Liver Function Test, Renal Function Test and urine analysis were all within normal limits. Enzyme-Linked Immunosorbent Assay (ELISA) for Human Immunodeficiency Virus was non-reactive. Serum Treponema pallidum hemagglutination assay (TPHA was negative). Rapid diagnostic rK39 was positive. Abdomen and pelvic ultrasonography showed normal scanning. Fine needle aspiration cytology from cervical lymph nodes did not reveal any L. D. bodies. Polymerase chain reaction (PCR) was not done due to unavailability in our institute.

Currently patient is under treatment following National guideline with improving health status [2].

DISCUSSION

There is paucity of published data on the clinical and epidemiological profiles of ML. It is caused primarily by Viannia subgenus species of Leishmania *(L. braziliensis, L. guyanensis, L. panamensis)* and *L. amazonensis* in Central and South America. The highest risk of ML occurs in the Bolivia (90%), Peru and Paraguay. Globalization, international travel and migration have increased the prevalence of Leishmaniasis in worldwide countries. Poverty, malnutrition, poor hygiene, environmental and climate change or an immunocompromised state are the potential risk factors [3].

ML is considered as metastatic complication of CL in less than 5% cases and develops either concurrently or years to decades after the clearance of cutaneous lesions [1]. CL usually heal on their own even without treatment leaving ugly scars [4]. Localization of cutaneous lesions on the upper half of the body has been reported as a risk factor for ML [5, 6]. Unlike cutaneous lesions, ML does not heal spontaneously. Parasite disseminates from amastigotes on the skin through direct extension to adjacent mucosa or the hematogenous or lymphatic system [1].

Lesions are usually described as whitish, red nodules or polypoidal masses, developing in a swollen mucosa of nose and mouth with oropharynx [7]. Lesions are characterized by mucosal destruction. Initial and prominent features include nasal systems which may progress to mucosal destruction of naso-oropharynx and larynx with complaints of disfigurement, aspiration, bleeding or fatal respiratory compromise [8].

Parasite and host immune factors greatly influence the clinical syndrome and severity of infection. The diagnosis is often challenging as it mimics many other infectious or malignant disease [9].

Multiple diagnostic testing methods should be performed to maximize diagnostic yield in the endemic regions. Definite diagnosis requires identification of parasite in histology, aspiration/ touch smear, culture or molecular analysis via PCR. Of these PCR is the most sensitive test, particularly in paucicellular yield histopathology or aspiration smear [10]. In Hematoxylin & Eosin or Giemsa stain, visualization of amastigotes in histiocytes as intracytoplasmic, small spherical to ovoid and measures 1-5μm is called as L. D. Bodies. They possess a large nucleus and prominent kinetoplast which is important for definite diagnostic purpose [11]. In resource poor setting Dermoscopy, Serological test (Direct Antiglobulin Test, Immunofluorescence, ELISA, Western blot analysis), rapid diagnostic rK39 test are other useful diagnostic aids [12].

For prevention, unfortunately, options like vaccination and territory control both are still largely unsatisfactory [7]. Its management can be complicated due to diagnostic delay, low indices of suspicion among health workers, low sensitivity of diagnostic tests, poor access to molecular testing, limited treatment options, and adverse treatment effects [12].

CONCLUSION

In our case, severity of symptom associated with progression of oropharyngeal lesion misled the clinical suspicion for oropharyngeal carcinoma. As epidemiology of leishmaniasis is changing, it should be considered as one of the differential diagnosis in endemic and nearby endemic zone. Screening examination of mucosal lesions should be recommended in all the patients diagnosed with CL. Furthermore, taking into consideration of all the discussed multi factors, elimination of the disease is still a challenge for our health community.

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