

# VISCERAL LEISHMANIASIS (KALA-AZAR) WITH A RARE MANIFESTATION OF SUBMENTAL ADENOPATHY

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## ABSTRACT

Visceral leishmaniasis is a protozoar disease caused by *L. donovani*. Transmission to humans (incidental host) occurs via insect bite and the phlebotomous (female sand-fly) transmits the disease.

Common presentations of the disease include hepatomegaly and splenomegaly, but fever, chills and night sweats may be part of the disease's symptoms. Lymphadenopathy without systemic manifestations is a very rare presentation of the disease.

We hereby present a 9 year old boy in whom the only presentation of visceral leishmaniasis in him was a 2×1.5cm submental mass without any signs or symptoms. After excisional biopsy Leishmaniasis was confirmed and the patient underwent medical management that resulted in complete recovery.

Visceral leishmaniasis must be considered as a cause of lymphadenopathy in endemic areas.

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## INTRODUCTION

Visceral leishmaniasis has re-emerged from near eradication in Iran. The annual estimate for the incidence and prevalence of this disease is 0.5 per million and 2.5 per million respectively.<sup>1</sup>

The term "Leishmaniasis" refers to a spectrum of diseases caused by the protozoar *Leishmania*.

There are three different forms of leishmaniasis:

- 1) Cutaneous L.; caused by *L. tropica*
- 2) Mucocutaneous L.; caused by *L. braziliensis*
- 3) Visceral L.; caused by *L. donovani*<sup>2</sup>

In Iran visceral leishmaniasis is endemic in rural areas and most of the time occurs in young children before age 10.<sup>1,3</sup>

It is a vector-borne zoonosis disease with common reservoirs being rodents, small mammals and canines,

but human is an incidental host.

Transmission to humans occurs via insect bite, where phlebotomous (female sand-fly) transmits the disease. These flies are often active in tropical zones and can not be found during the cold months. They are immobile in daytime and their activity is the most during early hours of night.<sup>3</sup>

## CASE REPORT

A 9 year old boy, an inhabitant of Bajegan (a region in the Bafgh district) referred to the Otolaryngology-Head and Neck Surgery clinic due to a submental mass.

The patient developed this mass 2 months ago, which enlarged in size gradually. No fever, chills, lack of appetite or local pain was associated with this mass.

The patient had received no medical therapy since that time.

In physical examination the only clinical finding was a 2×1.5cm mass which was completely mobile without local tenderness or signs of inflammation. There was no abnormal finding in other organs.

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## Kala-Azar with Submental Adenopathy

The patient was hospitalized and FNA and excisional biopsy was accomplished. The result of cytology and pathologic study confirmed the diagnosis of visceral leishmaniasis.

He received treatment after consultation with an infectious disease specialist and recovered completely.

### DISCUSSION

Leishmania lives as an intracellular parasite in reticuloendothelial cells, monocytes, polymorphonuclear cells and other phagocytic cells. This parasite multiplies via binary division in the cell and then the cell ruptures. Thereafter the parasite infects other cells and this cycle continues.

Sandflies ingest amastigotes containing macrophages when they take a blood meal. Amastigotes transform to promastigotes (flagellated form) which then multiply and differentiate in the sandfly gut.

The life cycle will be completed approximately 1 week later when metacyclic promastigotes are deposited as the sandfly attempts to take its next meal.<sup>4</sup>

The main pathological finding is multiplication of phagocytic cells of the reticuloendothelial system, in which significant numbers of leishmania are present. Most of the changes occur in the liver, spleen and bone marrow.

In lymph nodes the pathological picture may come to resemble necrotizing and suppurative lymphadenitis and have to be distinguished from other causes of necrotic suppuration such as tuberculosis and cat-scratch disease.<sup>5</sup>

The incubation period of the disease ranges from 4-6 months.<sup>6</sup>

Primary skin involvement is rarely noticed. The onset of visceral leishmaniasis can be silent and insidious without detectable fever.<sup>7</sup> In some cases however, there is an abrupt onset of fever, chills, night sweats and vomiting. Hepatomegaly and splenomegaly are the hallmark of visceral leishmaniasis.

Lymphadenopathy without systemic manifestations or skin involvement is an extraordinary form of visceral leishmaniasis.<sup>8,9</sup>

Rarely a localized lymphadenopathy may be the only manifestation of the disease. This shows that clinical findings vary by the geographic areas where the disease is seen. In the Mediterranean, Malta, South Africa and India, lymphadenopathy has been a major manifestation of the disease.<sup>1,10</sup>

Laboratory findings include leukopenia, thrombocytopenia and anemia; among them leukopenia is the most significant.

Hypergammaglobulinemia is common, erythrocyte sedimentation rate is elevated and coagulation tests may

be impaired.<sup>2</sup> Formol gel test is usually positive.<sup>9</sup>

The diagnosis is confirmed by identifying leishmania amastigotes in tissue or by growing promastigotes in culture (from samples of spleen, bone marrow, liver or lymph node aspiration) or serologic exams like complement fixation test (CF), indirect immunofluorescence assay, hemagglutinin test and ELISA technique.<sup>3,11</sup>

The leishmania skin test, also known as the Montenegro test indicates delayed-type hypersensitivity, and has a low value. This test is negative in persons with visceral leishmaniasis but it becomes positive in the majority of those who undergo successful medical therapy and in those with self-resolving infection.<sup>3</sup>

The treatment of choice is Glucantime, prescribed intramuscularly with a dose of 20 mg/kg daily for 3-4 weeks.<sup>2</sup>

Patients who undergo successful medical therapy must be examined again at 1 month intervals for 6 months and subsequently at one year. At each visit, the spleen must be examined, CBC and IgG level must be determined and tissue cultures checked for leishmania.<sup>2</sup>

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