

Cytodiagnosis of Visceral Leishmaniasis in Lymph Node Aspirate: A Rare Case Report

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ABSTRACT

Leishmaniasis is a parasitic disease caused by leishmania species. There are three main forms of the disease: Cutaneous, Mucocutaneous and Visceral Leishmaniasis (VL). VL with lymphadenitis is an uncommon presentation. Visceral leishmaniasis is usually diagnosed by identification of parasite Leishman Donovan (LD) bodies in bone marrow aspirates but cytological diagnosis of VL on lymph node aspirates has rarely been reported. This is a case report of a 37-year-old male presenting with multiple isolated cervical lymphadenopathy for three months. The lymphadenopathy was initially suspected to be tubercular in nature which is highly prevalent in India and lymphadenopathy is its common presenting feature, but on Fine Needle Aspiration Cytology (FNAC) of the node, multiple leishmania parasites were identified. Lymphadenopathy due to leishmaniasis is rare in India and most of the previous cases reported have association of leishmaniasis with Human Immunodeficiency Virus (HIV) infection. But this case is unusual as it is not associated with HIV infection. Thus, leishmaniasis should be included in the differential diagnosis of lymphadenopathy, especially in patients coming from endemic region. Cytology can be very helpful in such cases for early diagnosis and further management.

Keywords: Cytology, Leishman donovan bodies, Leishmanial lymphadenitis, Parasite

CASE REPORT

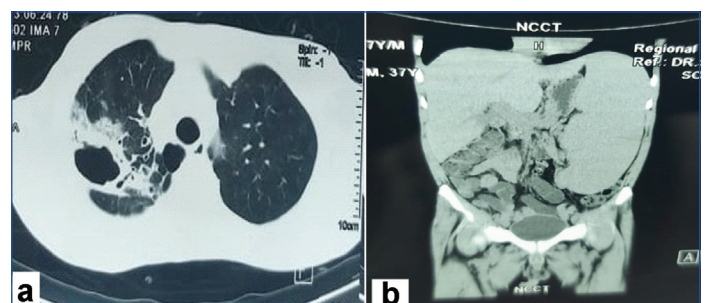
A 37-year-old male from Bihar came to Medicine Outpatient Department (OPD), presenting with painless, swelling in left cervical region for past three months duration which gradually increased in size. He also had history of low-grade intermittent fever, mild pain in abdomen on and off, anorexia and weakness for past three months. He had a past history of pulmonary Tuberculosis (TB) with completed treatment five years back. General physical examination revealed enlarged bilateral cervical lymph nodes measuring 0.5-2 cm in size, non tender and non matted.

On palpation, moderate hepatomegaly and massive splenomegaly was also noted [Table/Fig-1]. Investigations revealed few abnormal parameters like haemoglobin 6.4 g/dL (normal range 13-16 g/dL), total leucocyte count 1700/ μ L (normal range 4000-11000/ μ L) with P26L65M08E01 and platelets 1 lac/ μ L. (normal range 1.5-4 lac/ μ L). Routine biochemical investigations were within normal limits. Serology for HIV and Hepatitis B Virus (HBV) by Enzyme-linked Immunosorbent

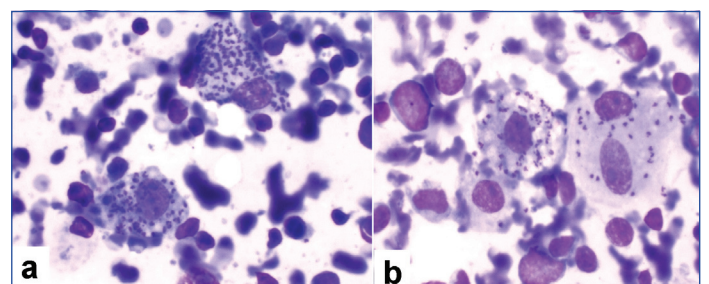
Assay (ELISA) was non reactive. Mantoux test was negative. Sputum was negative for acid-fast bacilli. Computed Tomography (CT) of the chest and abdomen examination revealed cavitary consolidated lesion in right upper lobe of lung, hepatomegaly and splenomegaly suggestive of disseminated tuberculosis [Table/Fig-2]. Based on these findings he was started again on antitubercular drugs. However, there was no improvement in the patient's overall symptoms. Then he was recommended for FNAC of cervical lymph nodes. FNAC was performed and Giemsa stained smears revealed a polymorphic population of cells composed of lymphocytes, histiocytes, and macrophages filled with leishmania amastigotes LD bodies [Table/Fig-3]. Ziehl-Neelsen staining for acid fast bacilli was negative. A final diagnosis of leishmania lymphadenitis was made. Patient refused treatment and went back to his native state and was lost to follow-up.



[Table/Fig-1]: Photograph of patient's abdomen with marked areas showing hepatomegaly and splenomegaly.



[Table/Fig-2]: a) CT scan of chest showing cavitary consolidated lesion in right lung; b) CT scan of abdomen showing hepatomegaly and splenomegaly.



[Table/Fig-3]: a) Cytological examination of lymph node aspirate showing polymorphous population of lymphoid cells and macrophages filled with intracellular leishmania amastigotes (LD bodies) (Giemsa stain, x100); b) Safety pin shaped LD bodies with kinetoplast within macrophages (Giemsa stain, x100).

| Study | Year | Place | Age (years)/sex | Clinical presentation | HIV status | Lymph node involved | Bone marrow findings |
|-----------------------------|------|-------------|-----------------|--------------------------------------|------------|---------------------------------|----------------------|
| Yaduvanshi A et al., [8] | 1999 | New Delhi | 30/M | Fever with pancytopenia | Positive | Inguinal | Absent |
| Sharma M and Malhotra A [9] | 2012 | West Bengal | 28/F | Asymptomatic | Negative | Inguinal | Absent |
| Nandi M et al., [10] | 2012 | West Bengal | 6/M | Fever | Positive | Bilateral cervical | Negative for LD body |
| Shelkar R et al., [11] | 2014 | Maharashtra | 27/F | Pancytopenia | Positive | Left cervical | Positive for LD body |
| Chaudhary NS et al., [12] | 2015 | Gurgaon | 41/M | Fever with pancytopenia | Positive | Mediastinal | Not done |
| Bode AN et al., [7] | 2015 | Maharashtra | 26/F | Fever with pancytopenia | Positive | Left cervical | Positive for LD body |
| Agarwal P et al., [13] | 2017 | New Delhi | 42/M | Fever with pancytopenia | Positive | Left submandibular and axillary | Positive for LD body |
| Parmar P et al., [14] | 2018 | Rajasthan | 18/M | Asymptomatic | Negative | Left arm | Positive for LD body |
| Pal S et al., [15] | 2018 | West Bengal | 38/M | Fever with weight loss | Positive | Right epitrochlear and cervical | Positive for LD body |
| Rana SS et al., [16] | 2021 | Chandigarh | 34/M | Malaise, fever, weight loss | Negative | Mediastinal | Not done |
| Present case | 2022 | Haryana | 37/M | Pancytopenia with hepatosplenomegaly | Negative | Bilateral cervical | Not done |

[Table/Fig-4]: Reported Cases of Visceral leishmaniasis reported on Lymph Node FNAC in India [7-16].

DISCUSSION

Leishmaniasis also called as kala-azar is a protozoan disease caused by different species of leishmania which is an obligate intracellular parasite and transmitted by bite of infected female sand fly. The amastigote forms of leishmania species spread through haematogenous route in the human body and affect the macrophages of mononuclear phagocyte system [1]. It's annual incidence rate is 0.5 million in the world but the infection is endemic in eastern India mainly Bihar, West Bengal and Uttar Pradesh [2]. It is characterised into three main types-visceral, cutaneous and mucosal [1]. Visceral Leishmaniasis (VL) is usually diagnosed by identification of parasite Leishman Donovan (LD) bodies in bone marrow, spleen and liver aspirates. Other methods such as tissue biopsy, immunohistochemistry and molecular methods are also used. However, lymph nodes involvement in kala-azar with demonstration of LD bodies by Fine-Needle Aspiration Cytology (FNAC) has rarely been reported in VL [3]. Early accurate diagnosis and treatment are important, otherwise it can be life threatening. This is a rare case report of VL involving cervical lymph node and diagnosed on cytology. Life cycle of leishmaniasis has two forms: the promastigote form which replicates in sandfly and is extra cellular and the intracellular amastigote form which multiplies in mammalian host [4]. The different clinical manifestations depend upon infection by different species of the parasite and the host immune response [1,3]. Cutaneous leishmaniasis is characterised by papular, nodular and ulcerative skin lesion, whereas in mucocutaneous/mucosal leishmaniasis there is involvement of mucosa of the nose and throat [5]. Visceral leishmaniasis is the most severe type caused by *L. infantum* and *L. donovani* presents with fever, anaemia, weight loss, darkening of skin and hepatosplenomegaly [6]. The diagnosis of leishmaniasis is mainly based on detection of characteristic amastigote forms also known as LD bodies in microscopic examination of bone marrow, spleen and lymph node aspirates [1]. Microscopically, LD bodies appear as small round/ovoid structures of 2-5 µm (diameter) with fine membrane and central round nucleus and rod shaped kinetoplast [7]. Although, it is an endemic disease in eastern India but peripheral lymphadenopathy in Indian VL or kala-azar is a rare finding. It is an incidental finding on cytology with only a few cases reported till date where leishmaniasis has been mainly diagnosed on FNAC.

Most of the previous cases reported have association of leishmaniasis coexistent with HIV infection (an immunocompromised state) [Table/Fig-4] [7-16]. But this case is rare as it is not associated with HIV infection. The cases of leishmaniasis reported in non endemic regions could be due to migrant population from endemic regions [13]. The diagnosis of leishmaniasis is often difficult due to lack of awareness, non specific symptoms similar to other commonly occurring diseases, such as malaria, typhoid and TB;

and microscopically difficult identification of amastigotes in bone marrow smears [7]. Cervical lymph adenopathy includes a variety of diseases in the differential diagnosis out of which tuberculosis, fungal infections, bacterial lymphadenitis and lymphomas are most common. Our patient had cervical lymphadenopathy with and hepatosplenomegaly as the predominant presentation. So, diseases like disseminated tuberculosis and lymphoproliferative disorders were included in the differential diagnosis. Leishmaniasis was not included in the differentials because lymphadenopathy is a rare finding in Indian VL, and can closely mimic tuberculosis which is highly prevalent in India and lymphadenopathy is its common presenting feature. A clinical diagnosis of TB was made based on clinical and radiological findings and Antitubercular Therapy (ATT) was started, but due to non improvement of symptoms, patient was sent for FNAC to rule out lymphoproliferative disorders. However, the cytology smears revealed multiple macrophages filled with LD bodies and diagnosis of leishmanial lymphadenitis was made. So, FNA cytology is a quick, reliable and a definitive diagnostic method and should always be considered as a method of choice in lymph node enlargement.

CONCLUSION(S)

The index case was clinically challenging with past history of TB and ATT twice and presenting with intermittent fever, lung cavity and peripheral lymph adenopathy. Although, leishmaniasis is a rare cause of lymph node enlargement, it should be considered as a differential diagnosis of all patients from endemic areas. FNAC is an excellent tool for the early diagnosis of leishmanial lymphadenitis.

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