

CASE REPORT

Visceral Leishmaniasis with Generalized Involvement of Lymph Nodes in a 55-Year-Old Woman: A Case Report

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Abstract

A 55-year-old woman was presented with fever, splenomegaly, generalized lymphadenopathy and weight loss. The patient came from Sreepur, an endemic region of Kala-azar in Bangladesh. About 2 years back, the patient presented with fever and splenomegaly and on investigation, ICT for Kala-azar was found positive. Thus the patient was diagnosed as a case of visceral Leishmaniasis and was treated with Miltefosin in full dosage form. The condition was improved with the treatment. After about a year of treatment the patient again developed fever and splenomegaly; this time was also generalized lymphadenopathy. Lymph node biopsy showed non-Hodgkin's lymphoma and bone marrow study showed the presence of LD-bodies. FNAC from cervical and epitrochlear lymph nodes showed the presence of LD bodies without the presence of any malignant cells, so she was diagnosed as a case of treatment failure of visceral Leishmaniasis with lymphatic involvement. [J Sci Found, 2012;10(2):80-84]

Keywords: Non-Hodgkin's lymphoma, Visceral Leishmaniasis, Kala-azar, treatment failure

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Introduction

Leishmaniasis is an intracellular infection caused by the protozoon *Leishmania* (Guerin et al., 2002). Rodents and canines are the normal reservoirs; the vector is the sand fly and humans are the host (Chappuis et al., 2007). The infection can provoke one of three clinical forms known as cutaneous, visceral, or mucocutaneous depending on the *Leishmania* species and the host immune response (Matlashewski et al., 2011). Leishmaniasis is endemic in about 90

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countries; at least 12 million people are reported to be infected worldwide, and 400,000 new cases occur each year (Srivastava et al., 2011). More than 90% of all cases of VL occur in Bangladesh, India, Nepal, Sudan, and Brazil (Argaw et al., 2013). VL is a systemic disease characterized by fever, anemia, leukopenia, hepatosplenomegaly, and weight loss that typically develop several months or even years after the initial infection (Mondal et al., 2013). But lymph node involvement in Leishmaniasis is rare in this sub-continent, although manifestation of leishmaniasis in the form of superficial lymph node enlargement is known to occur in the Mediterranean countries, Africa and China (Amin et al., 2013). In this report, the case of a patient of Visceral Leishmaniasis (VL) who did not respond to treatment with miltefosine was developed an unusual manifestation of generalized lymphadenopathy. The case was diagnosed on the basis of presence of *Leishmania donovani* (LD) body in fine needle aspiration cytology (FNAC) of lymph nodes and in bone marrow aspiration.

Case Presentation

A 55-year-old housewife living in Sreepur under Mymensingh district, an area where Leishmaniasis is endemic was presented with prolonged, low grade fever and splenomegaly for 2 months, weight loss for 2 years and generalized lymphadenopathy. The patient has a history of similar type of illness about 2 years back. On that occasion the patient presented with prolonged fever and splenomegaly and severe anemia; however there was no lymphadenopathy. There was associated cough with sputum production. The complete blood count (CBC) revealed Hemoglobin (Hb) 5.3 gm/dL, ESR 160 mm in 1st hour, red blood cells $1.8 \times 10^{12}/L$, white blood cells $3.50 \times 10^9/L$, neutrophils 46%, lymphocytes 49.0%, monocytes 4%, eosinophils 1%, basophils 0%, platelets $100 \times 10^9/L$. Chest-x-ray showed bilateral pneumonia. Sputum was negative for AFB. USG of whole abdomen showed splenomegaly and enlarged lymph nodes in para-aortic region. Serum creatinine level was 0.8 mg/dL and urinary total protein was within normal range. Blood was sent for culture and sensitivity; however, there was no growth. ICT for Kala-azar was done and was found positive. Therefore the initial investigation reports were consistent with Kala-azar and the patient was treated with tablet Miltefosin for 1 month. With this treatment the physical condition was improved and was remained afebrile for about a year. Now after 11 months of completion of the treatment the patient again presented with fever and a lump in left upper abdomen. The patient also complained of undocumented; however, significant weight loss was noticed for last two years. Fever was low-grade and continuous in nature which subsided after taking antipyretic drug, though recurred again. There is no evening rise of temperature and no history of night sweats. The mass in her abdomen is gradually increasing in size. On examination, she was febrile and pale and multiple superficial lymph nodes were palpable on cervical, epitrochlear and inguinal region of both sides. The lymph nodes varied in size and shape as well as consistency was firm; some were matted and are freely mobile. Liver was 5 cm palpable below the costal margin and spleen was increased in size, 17 cm along its long axis. Other clinical findings were normal. This time CBC report showed Hb 7.4 gm/dl, ESR 140 mm in 1st hour, red blood cells $2.7 \times 10^{12}/L$, white blood cells $2.3 \times 10^9/L$, neutrophils 36%, lymphocytes 58%, monocytes 4%, eosinophils 2%, basophils 0%, platelets $120 \times 10^9/L$. Chest-x-ray showed dense opacity in the mid zone of right lung. USG of the whole abdomen showed splenomegaly and enlarged perihilar lymph nodes. The patient was positive for HBsAg and negative for HCV; prothrombin time was 15.7 seconds and INR was 1.31, SGPT was 23 U/L, LDH was 425 U/L, serum bilirubin was 0.5 mg/dl, total protein was 120 g/L and serum albumin was 23 g/L and upper GIT endoscopy revealed no abnormality. The routine urine examination was normal and serum creatinine was 1.2 gm/dl. Excisional biopsy was done from right cervical lymph node and the report was non-Hodgkin's lymphoma

(intermediate grade). Bone marrow study was done and it showed many intracellular and extracellular LD-bodies and plasma cells and histiocytes were increased. Lymph node biopsy report was reviewed again and it showed angio-immunoblastic T-cell lymphoma. Then a fine needle aspiration cytology (FNAC) was done from cervical and epitrochlear lymph nodes that revealed the presence of LD-bodies without malignant cells. Therefore she was diagnosed as a case of treatment failure of visceral leishmaniasis with lymph nodes involvement and was treated with Amphotericin B. After treatment with Amphotericin B the patient became afebrile and the spleen also started to decrease in size.

Discussion

In Bangladesh the visceral Leishmaniasis is still alarming. Many untreated or incompletely treated cases develop many complications like PKDL, chronic infection etc., but progression to lymph node involvement is rare (Khan et al., 2014). In any patient with VL and generalized involvement of lymph nodes should be evaluated for other common causes of lymphadenopathy like tuberculosis, lymphoma, leprosy and fungal infection (Shamsuzzaman et al., 2000; Islam et al., 2002). It has been reported that there are 30-100 subclinical infections for every overt case of visceral leishmaniasis (Islam et al., 2004). There are many risk factors for the development of clinical diseases like malnutrition (Ho et al., 1982), immunosuppressive drugs, and, especially, HIV co-infection (Cerf et al., 1987; Alvar et al., 1997). The number of co-infections will continue to rise, notably in India and Brazil, where the urban HIV epidemic and the rural visceral leishmaniasis epidemic are increasingly coming into contact. Cases of co-infection are seen as an imported disease in non-endemic areas (Berman 1997). Co-infected patients may be difficult to diagnose, respond poorly to treatment, and relapse repeatedly (Desjeux 1997).

In this present case report non-Hodgkin's lymphoma was diagnosed. Visceral leishmaniasis may be co-existent with carcinogenic conditions like NHL; therefore carcinogenic conditions should be excluded in patients suffering from VL like infectious diseases so here we had to exclude lymphoma by lymph node biopsy and FNAC from lymph nodes. The lymph node biopsy taken in the first occasion referred to an atypical form of NHL but later on FNAC of the lymph nodes revealed the presence of LD-bodies but no malignant cells. It is very rare indeed. Co-morbidity with cancer among the visceral Leishmaniasis patients is very uncommon; therefore the present case report has given a new clinical array to leishmaniasis infection.

Drugs currently available for the treatment of KA include miltefosine, amphotericin B-deoxycholate, liposomal amphotericin B, other lipid formulations, and paromomycin (Pintado and Lopez-Velez 2001). Miltefosine is the first-line drug, and amphotericin B-deoxycholate is used as a second-line drug for treatment of KA (Guerin et al., 2002). This patient was treated with Miltefosin in full dosage form earlier. However she has developed Kala-azar again in less than one year which is known as relapse case of Kala azar (Rai et al., 2013). VL relapses are rare in otherwise healthy subjects but frequent in immunodeficient patients. So it can be a case resistant to Miltefosin. Because of increasing resistance to such other agents, Amphotericin B is increasingly used to treat VL and is currently the first-line therapy for immune-deficient patients (Pintado and Lopez-Velez 2001). Efficacy seems to be similar with standard and liposomal Amphotericin B, and the choice between the different preparations is often dictated by availability and cost. Here this patient was treated with Amphotericin B after treatment failure with miltefosine and the response was good within 5

days. Therefore it is very important to find out the associated co-morbidities of the kala-azar patients during diagnosis.

Conclusion

It can be concluded that lymphatic leishmaniasis as a differential diagnosis needs to be kept in mind when a patient with generalized lymphadenopathy is encountered from an area endemic for Kala-azar.

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