Kala-azar without splenomegaly: A rare presentation

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ABSTRACT

Visceral leishmaniasis is a major public health problem in Bangladesh, North East India, Nepal, Sudan and North East Brazil. In India, leishmaniasis is more prevalent in Bihar, Jharkhand, West Bengal and Uttar Pradesh. We present the case of a 55-year-old male farmer from Himachal Pradesh with complaints of fever for 2.5 months, appetite loss and weight loss for 1 month. On evaluation, he was found to have pancytopenia, transaminitis, and hyperbilirubinemia. Tropical fever serology and viral markers were negative. Blood and urine cultures were sterile, and ascitic fluid was acellular and high SAAG with normal ADA. Bone marrow was done due to non-responding pancytopenia which reveals intracellular amastigote form of Leishmania Donovani. A final diagnosis of Kala-azar without splenomegaly with moderate ascites was made as an absence of splenomegaly was the most striking aspect in our patient.

Keywords: *Atypical presentation, Kala Azar, Leishmania donovan bodies, Splenomegaly.*

Leishmaniasis is a vector-borne zoonosis having a variable clinical presentation in the form of visceral, cutaneous and mucocutaneous types depending upon the Leishmania species and immune responses of the hosts [1]. The disease mainly affects poor people in Africa, Asia and Latin America, and is associated with malnutrition, population displacement, poor housing, weak immune system and lack of resources. Out of 200 countries and territories reporting to the World Health Organisation (WHO), 97 countries and territories are endemic for leishmaniasis in 2017 [1]. Leishmaniasis is still a public health problem in several underdeveloped countries.

Progressive leukopenia and anemia are striking features of Leishmania donovani infection, Visualization of splenic amastigotes [Leishman-Donovan (LD) bodies] splenic aspirate is the most specific test. A rapid dipstick test based on the recombinant K39 protein is widely used for the rapid diagnosis of visceral leishmaniasis (VL). Liposomal amphotericin B is the optimal treatment for visceral leishmaniasis [2].

In the endemic area, subclinical forms of Kala-azar remain undiagnosed and can become a clinical challenge to the treating physician. We are presenting the case report of a 55-year-old male from an endemic area of Kala-azar with an atypical presentation. The rationale of this case report is to inform about the missing and under diagnosing Kala-azar due to its atypical presentation

CASE REPORT

A55-year-old male farmer from Himachal Pradesh, India, presented to our hospital with a history of intermittent fever for two and a half months, appetite loss and weight loss for1 month. There were no other significant localizing symptoms.

General physical examination reveals significant pallor, bilateral pedal edema, and abdominal distension. The vitals were stable and there was no lymphadenopathy. Systemic examination reveals hepatomegaly (firm, non-tender) 3 cm below costal margin but there was no splenomegaly.

Hematological parameters show pancytopenia (with normal mean corpuscular volume and reticulocyte count). At admission, the hemoglobin was 7.5 gm%, total leukocyte count (TLC) was 900/mm^3, differential leukocyte count (DLC) was not possible and platelet count 11000/mm^3. Peripheral blood film shows microcytes, macrocytes, anisocytosis with reduced platelets on the smear. Lactate dehydrogenase (LDH) and renal function tests were normal. The liver function tests showed total bilirubin 1.9 mg%, aspartate aminotransferase (AST) 65 IU/L, alanine aminotransferase (ALT) 38 IU/L, alkaline phosphatase 936U/L, total protein 6.2gm/dl and albumin 3.0 gm/dl.

Peripheral smear for microfilaria and malaria were negative. Viral markers (HBV, HCV, HIV) and other tropical fever serology (dengue, scrub, malaria) were negative. Chest X-ray and work up for pyrexia of unknown origin (PUO) was normal, blood and urine cultures were sterile. Workup for hemolytic anemia was negative. Ultrasound (USG) whole abdomen showed liver span 17 cm with normal echotexture without splenomegaly with mesenteric lymphadenopathy and moderate ascites. The ascitic fluid was a cellular with glucose 141 mg/dl, protein 1.4 gm/dl, albumin 0.5gm/dl and adenosine deaminase (ADA) was 14 IU/L. The culture was sterile and there was no evidence of malignant cell and acid-fast bacilli on ascetic fluid cytology. No other radiological, biochemical, cytological or serological evidence of tuberculosis or malignancy found.

Bone marrow aspirate examination was performed in view of non-responded pancytopenia which revealed intracellular

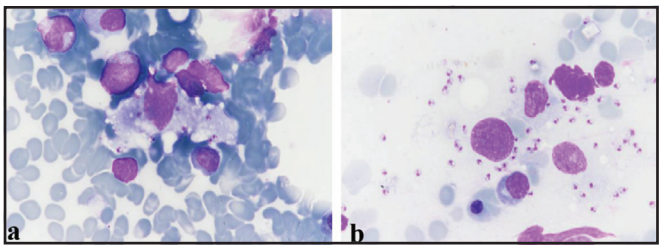

Figure 1: (a) Intracellular and (b) extracellular amastigote form of Leishmania donovani.

amastigote form of LD in macrophages (Fig. 1). A final diagnosis of VL (Kala-azar) was made as there was an absence of splenomegaly with moderate ascites. Later on, the diagnosis of VL was made and intravenous (IV) liposomal amphotericin was started, continued for a period of 2 weeks. The patient responded for therapy, fever subsides, his general condition was start improving, appetite increased and hematological parameters were also improved. The patient was completely recovered over a period of 2 months.

DISCUSSION

Kala azar has variable clinical presentation ranging from asymptomatic to full blown disease. Leishmaniasis not always cause clinical disease as asymptomatic and subclinical forms are more frequent. The classical presentation of Kala-azar includes fever, asthenia, weight loss, anemia, splenomegaly, hepatomegaly and sometimesadenopathy. Splenomegaly appears early and increases gradually in relation to the duration of disease [1]. The presence of amastigote form in the macrophages remains the hallmark of Leishmania infection.

Badaro et al. [3] characterize the subclinical form of VL as mild and non-specific clinical manifestation lasting for more than 3 weeks; however in our case, clinical features persist for a period of 3 months. Sameer et al. [1] also found an atypical presentation of Kala-azar without splenomegaly with evidence of hemolytic anemia, while in our case, the patient was having pancytopenia probably due to bone marrow suppression. Some other studies like Gawade et al [4] present a case of VL in which progressive weakness abdominal discomfort and loss of appetite last for 6 months followed by petechial hemorrhage over the body, while in our case, hemorrhagic manifestation was not seen.

Kumar R et al [2] presented a case series of nine cases; out of them, two patients were having portal hypertension, five patients had no evidence of splenomegaly and one patient was having gastrointestinal symptom in which endoscopic biopsy detected LD bodies on histopathological examination while in our case, the patient was not having portal hypertension. Ravishankar SN.et al [5] presented a case of VL with an absence of splenomegaly and association with hepatocellular carcinoma. Mohan A et al [6] presented a case report on VL without fever or splenomegaly in an elderly person presenting the only clinical feature of anemia. Geraci JE et al [7] presented a case of VL as a cause of fever of unknown origin in which the diagnosis was made by exploratory laparotomy and splenectomy after diagnostic studies had failed to reveal the cause of fever. The patient was cured with 6 days course of therapy with sodium antimony gluconate.

VL should be considered in the differential diagnosis of patients with obscure fever who have traveled in endemic areas. Kala-azar is more common as asymptomatic or subclinical form as compared to typical presentation in the non-endemic area like Solon Himachal Pradesh, clinically overlap with connective tissue disorder, tuberculosis or malignancy [8,9,10]. Therefore in the literature, a few numbers of case reports on VL and its atypical presentation in various forms has been mentioned. The present case report contributes a similar kind of atypical presentation in the form of absence of splenomegaly.

CONCLUSION

In this case, the initial presentation was not typical of Kala-azar which later on diagnosed as visceral leishmaniasis. Unlike our case, none of the previously reported cases showed an overlap of tuberculosis with visceral leishmaniasis. Atypical features of Kala-azar are equally important, as long duration of fever without any typical feature should always be considered as a differential diagnosis of Kala-azar.

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