

Recurrent Kala Azar with Recurrent Post-Kala-Azar Anterior Uveitis in an Immuno-competent Child: A Case Report

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Received date: Nov 18, 2014, Accepted date: Dec 19, 2014, Published date: Dec 22, 2014

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Abstract

We are reporting a case of recurrent Kala azar with recurrent post-Kala-azar anterior uveitis in an 8 year old immune-competent child. Patient presented history of intermittent fever with loss of appetite and lassitude. Diagnosis of Kala azar was made on the basis of clinical examination and bone marrow microscopy. Child was treated with intravenous liposomal Amphotericin B, and was declared cured after 3 weeks. However, after one week of discharge, he presented with both eyes redness and on slit lamp examination bilateral anterior uveitis was detected. Uveitis was treated with topical steroids and cycloplegics. Relapse of Kala azar was noted 5 months after the first attack. He was treated with increased dose of intravenous liposomal Amphotericin B. After completion of treatment, bilateral anterior uveitis was noted. This was more severe than first attack, associated with fibrinous exudates in the left eye. Uveitis was successfully treated with topical steroids and cycloplegics. He presented with second relapse of Kala azar 7 months after the second attack and this time he was treated with intravenous liposomal Amphotericin B along with oral Miltefosine. On the 4th day of treatment, anterior chamber cells were noted bilaterally and this inflammation was controlled with topical steroids and cycloplegics.

Keywords: Kala azar; Uveitis; Recurrent; Amphotericin B

Introduction

Kala azar, the Indian name for visceral leishmaniasis (VL) is a protozoan parasitic disease caused by *L. donovani*. India contributes the highest number of VL cases worldwide, of which more than three fourth are reported in Bihar state. After proper drug treatment in immune-competent individuals, an effective life-long cellular immune response normally develops, and growths of residual parasites are suppressed [1]. However relapse of the disease does occur in a proportion of immune-competent patients, generally within 6-12 months of initial treatment despite negative end-of-treatment test-of-cure results [2].

We report a case of recurrent Kala azar with recurrent post-Kala azar anterior uveitis.

Case Report

A 8 year-old boy presented to us in July 2011 with a 6 weeks history of intermittent, moderate grade fever with loss of appetite and lassitude. Physical examination revealed pallor, with temperature of 100.2°F. Liver and spleen were palpable 3 cm and 5 cm below the right costal margin and the left costal margin, respectively. Malaria, Typhoid, and Tuberculosis were excluded by appropriate tests. Peripheral smear showed normocytic, hypochromic erythrocytes. Bone marrow tissue stained with Giemsa stain revealed LD bodies. Diagnosis of Kala azar was made and patient was started on intravenous 1 mg/kg Liposomal Amphotericin (AmBisome®) daily for 20 days. Patient became symptomatically better within 4 days after starting the treatment. After 20 days of treatment, patient was declared

to be cured with absence of LD bodies on bone marrow examination. After 1 week of discharge, patient came back to us with redness of both the eyes along with ocular pain and headache. Patient was evaluated in Ophthalmology department of our hospital. BCVA in both the eyes were 6/6. Marked circumciliary congestion was noted in both eyes, anterior chamber reaction was noted, with moderate number of tiny cells (20-25 in 1 mm × 1 mm slit beam) moving in the caloric stream, and a moderate flare (grade 2+). Absence of keratic precipitate (KPs) and hypopyon was noted. IOP in right eye was 16 and left eye was 14 mm Hg. Both eyes fundi were within normal limit. Patient was started on eye drops Prednisolone acetate 1%, 8 times a day, Moxifloxacin 0.5%, 3 times a day, and Homatropine 2%, 4 times a day. Within 10 days period, uveitis resolved, and steroid eye drop was tapered over 4 weeks period. Patient was apparently normal thereafter. However in December 2011, he again presented to us with moderate grade fever, lassitude and anorexia since 2 weeks duration. On examination, hepatosplenomegaly of same grade as in July month was noted (Figure 1A). Malaria, Typhoid, and Tuberculosis were excluded by appropriate tests. Microscopic examination of bone marrow tissue revealed LD bodies (Figure 1B) and diagnosis of relapse of Kala azar was made. ELISA test for HIV was negative. Intravenous AmbiSome was started in a dose of 1.5 mg/day and blood investigations were monitored routinely to rule out any toxicity of the drug. Patient started to respond with this treatment within a week time. AmbiSome was continued for total 3 weeks duration and at the end of 3 weeks, marrow examination didn't find any LD bodies. However, 7 days after completion of treatment, patient developed bilateral severe eye redness and intolerance to light. BCVA in both the eyes were 6/9, marked circumciliary congestion (Figure 1C). Uveitis was more severe compared to 1st attack with both eyes having AC cells 50-60 in 1 mm × 1 mm slit, fine KPs, with flare of 2+. Left eye had fibrinous exudates in AC (Figure 1C).

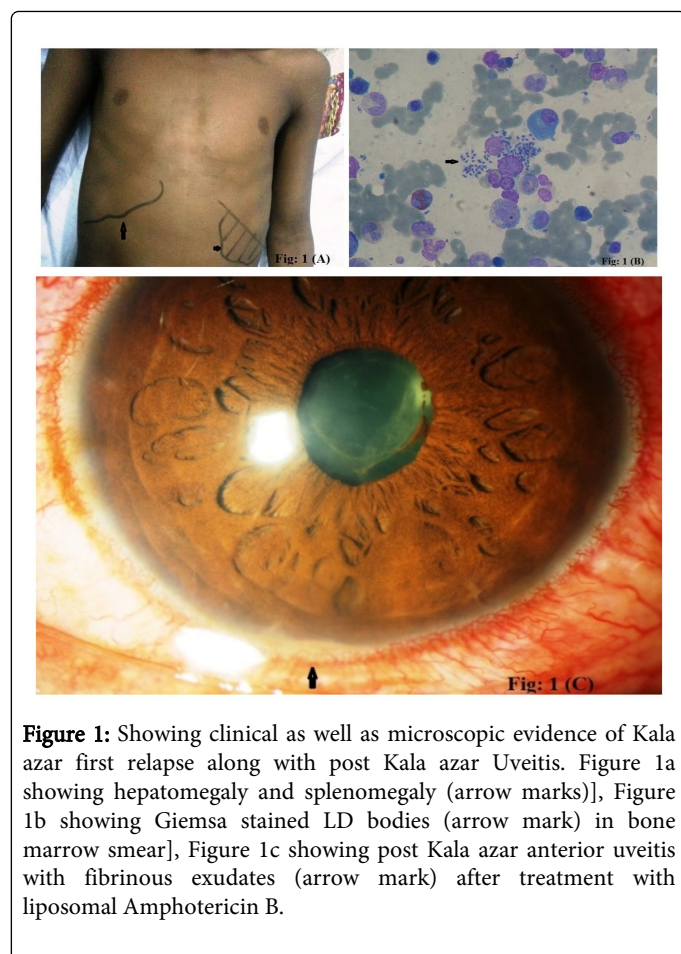


Figure 1: Showing clinical as well as microscopic evidence of Kala azar first relapse along with post Kala azar Uveitis. Figure 1a showing hepatomegaly and splenomegaly (arrow marks)], Figure 1b showing Giemsa stained LD bodies (arrow mark) in bone marrow smear], Figure 1c showing post Kala azar anterior uveitis with fibrinous exudates (arrow mark) after treatment with liposomal Amphotericin B.

Retrolental cells were absent in both the eyes and fundus was normal. Patient was started on eye drops Prednisolone acetate 1% every 1 hour, Moxifloxacin 0.5%, 3 times a day, and Homatropine 2%, 3 times a day and Atropine eye ointment 1% at night time. Uveitis resolved in 3 weeks period and steroid eye drop was tapered over 6 weeks period. In July 2012 patient again presented with relapse of Kala azar, with hepatosplenomegaly and LD bodies in bone marrow. Patient was started on oral Miltefosine (2.5 mg/kg/day) and Intravenous AmbiSome (1 mg/kg/day), and due to previous history of development of anterior uveitis, daily slit lamp examination was performed after starting the treatment. On 4th day, mild circumciliary congestion was noted with AC cells 10-15 in 1 mm × 1 mm beam. Patient was started on eye drops Prednisolone acetate 1%, 8 times in a day, Moxifloxacin 0.5%, 3 times a day, and Homatropine 2%, 4 times a day. This regimen of topical medications was continued till 4 weeks with IOP monitoring, there after tapered over 8 weeks period. AmbiSome was continued for a total of 3 weeks period and Miltefosine was continued for total of 4 weeks. At the end of 4 weeks, marrow tissue didn't reveal any LD body. Serological tests were performed to rule out any possible immunosuppressive disorder. Patient was also tested for HLA B-27, which was negative. Patient has been last seen in April 2014 and is free of any systemic or ocular problem.

Discussion

Ocular complications of kala azar are rare. The published literature on ocular complication of Leishmaniasis includes reports of retinal haemorrhage [3], interstitial keratitis [4], blepharoconjunctivitis [5], and destruction of intraocular tissue by the organisms [6]. Dechant et al. [7] in 1980 reported 3 cases of post Kala azar uveitis occurring during the course of, or shortly after the conclusion of the systemic illness. El-Hassan et al. [8] described bilateral anterior uveitis developing in 2 patients after successful treatment of Kala azar and Leishmania parasites were found in the iris tissue. Khalil et al. [9] have reported a case of blindness due to pan uveitis following visceral leishmaniasis.

Time relationship of the onset of the uveitis after starting the treatment in our case is more than coincidental. In our case 3 factors seem to be likely cause of this post Kala azar uveitis as Leishmania, Amphotericin B, and immune mediation. We presume the cause of uveitis due to alteration in cellular immunity, by a process of immune reconstitution, as described by Khalil et al. [10] for the development of post-Kala azar dermal leishmaniasis syndromes. The association of recurrent kala azar and post kala azar uveitis makes our case unique and timely recognition of this led to early initiation of treatment and can prevent the further sequelae.

References

1. Guerin PJ, Olliaro P, Sundar S, Boelaert M, Croft SL, et al. (2002) Visceral leishmaniasis: Current status of control, diagnosis, and treatment, and a proposed research and development agenda. *Lancet Infect Dis* 2: 494-501.
2. Burza S, Sinha PK, Mahajan R, Lima MA, Mitra G et al. (2014) Risk Factors for Visceral Leishmaniasis Relapse in Immunocompetent Patients following Treatment with 20 mg/kg Liposomal Amphotericin B (Ambisome) in Bihar, India. *PLoS Negl Trop Dis*. 8:e2536.
3. Montero JA, Ruiz-Moreno JM, Sanchis E (2003) Intraretinal hemorrhage associated with leishmaniasis. *Ophthalmic Surg Lasers Imaging* 34: 212-214.
4. Roizenblatt J (1979) Interstitial keratitis caused by American (mucocutaneous) leishmaniasis. *Am J Ophthalmol* 87: 175-179.
5. el Hassan AM, Khalil EA, el Sheikh EA, Zijlstra EE, Osman A, et al. (1998) Post kala-azar ocular leishmaniasis. *Trans R Soc Trop Med Hyg* 92: 177-179.
6. Kumar PV, Roozitalab MH, Lak P, Sadeghi El (1993) Ocular Leishmaniasis; A Cause of Blindness. *Iranian Journal of Medical Sciences* 18: 106-111.
7. Dechant W, Rees PH, Kager PA, Klauss V, Adalal H (1980) Post kala-azar uveitis. *The British Journal of Ophthalmology* 64: 680-683.
8. el-Hassan AM, el-Sheikh EA, Eltoum IA, Ghalib HW, Ali MS et al (1991) Post-kala-azar anterior uveitis: demonstration of Leishmania parasites in the lesion. *Trans R Soc Trop Med Hyg* 85: 471-473.
9. Khalil EA, Musa AM, Younis BM, Elfaki ME, Zijlstra EE et al. (2011) Blindness following visceral leishmaniasis: a neglected post-kala-azar complication. *Trop Doct* 41:139-40.
10. Khalil EA, Khidir SA, Musa AM, Musa BY, Elfaki ME et al. (2013) Post Kala-Azar Dermal Leishmaniasis: A Paradigm of Paradoxical Immune Reconstitution Syndrome in Non- HIV/AIDS Patients. *Journal of Tropical Medicine* 2013: 275253.