Gastrointestinal Leishmaniasis in Non-Endemic Region

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Abstract
We report a case of visceral leishmaniasis (VL) in an immunocompetent native from non-endemic region of India that presented with chronic diarrhoea. VL was not a differential diagnosis and was unexpectedly diagnosed as intestinal leishmaniasis through the identification of the Leishman-Donovan (LD) bodies in duodenal and colonic mucosa. The patient expired before receiving antileishmanial therapy.

Introduction
Visceral leishmaniasis is a disease of low altitude and is endemic in various parts of India, mainly Bihar, West Bengal and Orissa. However, epidemiological changes are taking place and an increased numbers of cases have been reported from natives of non-endemic areas like sub-Himalayan region.¹⁻³ Even with classical presentation the first diagnosis is never kala-azar in most cases. Atypical presentation of VL in a non-endemic area can lead to a diagnostic dilemma as index of suspicion is very low. The gastrointestinal involvement in visceral leishmaniasis is rare and has been reported either in those with concomitant HIV infection or in immunocompetent patients from endemic regions.⁴⁻⁵ This is a presentation of an unusual case of visceral leishmaniasis in a native of non-endemic region of India who presented with atypical clinical features, and was unexpectedly diagnosed as gastrointestinal leishmaniasis through the identification of the Leishmania organism in the unusual sites of duodenal and colonic mucosae. The patient had never visited any leishmania-endemic region in his life. We report this case for the following reasons: the patient belonged to non-endemic region, he presented with chronic diarrhoea, had apparently normal mucosa on endoscopy, LD bodies were found in duodenal and colonic mucosa on biopsy and he was immunocompetent.

Case Report
Fifty year male, agriculturist, smoker, nonalcoholic, native of Ravi river valley area situated at an altitude of 996 meters above the mean sea level of Himachal Pradesh, India was admitted in November 2014 with history of diarrhoea for 6 months. Patient used to pass five to six loose stools in a day. Stools were large volume without mucus and blood and contained undigested food. No history of pain abdomen was present. He gave history of intermittent fever for 6 months. Patient had no history of vomiting and loss of appetite. He reported loss of weight which was not documented. Review of other systems was normal. He had no significant past history. Treatment records revealed that the patient had received multiple courses of antibiotics from primary care physicians without any relief. He denied ever visiting any endemic area of visceral leishmaniasis. On examination, pallor was present and he was afebrile. No icterus and lymphadenopathy were present. His body mass index was 18 kg/m². Per abdomen examination, revealed massive splenomegaly (palpable 8 cms below left costal margin) and hepatomegaly. Rest of

Fig. 1: A) Duodenal biopsy showing normal villous:crypt ratio and granular clumps of LD bodies in the lamina propria (X 100, H & E); B) Duodenal biopsy under high power examination showing abundant macrophages with intracytoplasmic and extracellular bodies of leishmania (X 400, H & E); C) Oil immersion showing intracytoplasmic and extracellular LD bodies in duodenal biopsy (X 1000, Giemsa); D) Colonic biopsy showing macrophages with abundant intracytoplasmic and extracellular LD bodies (X 1000, H & E)

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the examination was normal. On investigations, hemoglobin was 7.5 gm %, leukocyte count was 2100/mm³ (neutrophils-45%, lymphocytes-46%) and platelet count was 1,62,000/mm³. Erythrocyte sedimentation rate was 60 mm in 1st hour. Microcytic hypochromic anemia was observed on peripheral smear. Blood glucose, renal function and liver function tests were normal. IgA tissue transglutaminase (TTG) and human immunodeficiency virus serology was negative. Stool examination was unremarkable. Chest X-ray was normal. Ultrasound abdomen showed hepatosplenomegaly and mesenteric lymphadenopathy. Upper gastrointestinal endoscopy was performed and found to be mostly normal except for antral gastritis and non-specific duodenitis. Biopsy was taken from the duodenum (D2) as a part of chronic diarrhoea investigation protocol. On colonoscopy multiple superficial small erythematous lesions in transverse colon were observed and a biopsy was taken from the site. Histopathological examination of duodenal biopsy revealed a well oriented adequate tissue with normal crypt villous ratio. Lamina propria showed granular clumps under low power (Figure 1A). High power examination showed abundant macrophages with intracytoplasmic and extracellular bodies of Leishmania in the lamina propria (Figure 1B, C).

Colonic biopsy showed focal clumps of macrophages with intracytoplasmic and extracellular Leishman Donovan bodies in lamina propria (Figure 1D). The patient expired before receiving antileishmanial therapy.

Discussion

Visceral leishmaniasis often presents with atypical features in the immunocompromised patient. The clinical picture in cases from non-endemic region is generally similar to that of already established in patients from endemic region of India but certain uncommon manifestations like leishmanial lymphadenopathy was reported by us in our previous study from non-endemic region.1 The gastrointestinal involvement in visceral leishmaniasis is rare and has been reported more frequently in those with concomitant HIV infection.4 Duodenum is the most common site though lesions have been observed from the oesophagus to the rectum. Prevalence of diarrhoea ranges from 5-26% in patients with VL, and presentation with chronic diarrhoea is rare.4 Diarrhoea was reported by 16.6% of patients in our previous study on VL.4 The exact pathogenesis of the diarrhoea is not clear. It is assumed that the symptoms in enteropathic visceral leishmaniasis may be a combination of the mechanical occlusion of the mucosa by parasites, bacterial overgrowth, partial villous atrophy, competition between the host and the parasite for nutrients, altered motility, bile salt deconjugation and lymphatic blockade.7

Conclusion

Clinicians and pathologists should be alert about the possibility of leishmaniasis in patients presenting with chronic diarrhoea from this non-endemic area. Initial failure to suspect visceral leishmaniasis might cause a diagnostic delay.

References