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Case Report

Lepromatous Leprosy with Type II Lepra Reaction and Jejuno-Jejunal Intussusception Possibly due to Strongyloidiasis and / or Mesenteric Lymphadenopathy Presenting as Acute Abdomen

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Strongyloides stercoralis can cause hyper-infection in immunosuppressed patients and can cause death if not treated. It often leads to missed or delayed diagnosis due to its nonspecific symptoms and signs. A 38-year-old male patient, a treated case of lepromatous (LL) Hansen's disease, presented with pain abdomen for four days, associated with vomiting, fever and melaena, and skin rash for three days. The skin rash was suggestive of erythema nodosum leprosum. He had been suffering from joint pains for the past four months, for which he had been using oral prednisolone 60 mg/day for four months. Stool examination showed *Strongyloides* larvae and occult blood. The contrast-enhanced computerized tomography of abdomen revealed jejuno-jejunal intussusception and mesenteric lymphadenopathy. Biopsy from duodenal mucosa revealed *Strongyloides* larvae and ova within the mucosal crypts. Immunosuppression secondary to chronic steroid intake, in this case, is the likely cause of strongyloidiasis hyper-infection. The association of LL and strongyloidiasis is infrequent. In leprosy under steroid therapy, the development of abdominal/pulmonary symptoms should raise a suspicion of stongyloidiasis. There is a need to study the incidence of strongyloidiasis in lepromatous leprosy cases to start blanket anthelminthic treatment as prophylaxis in case incidence is high.

Keywords : Leprosy, Immunosuppression, Strongyloidiasis, Mesenteric Lymphadenopathy, Intussusception.

Introduction

Leprosy is a chronic granulomatous infection caused by *Mycobacterium leprae,* principally affecting the peripheral nerves and skin. Type II lepra reaction can occur during the disease or even later after completing specific anti-leprosy therapy. Diagnosis of *Strongyloides stercoralis* hyper-infection in leprosy patients has been made rarely due to its nonspecific clinical features (De Souza et al 2014). In leprosy patients on

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prolonged steroid therapy, we should suspect *S. stercoralis* infestation in case of acute abdomen.

Case Report

A 38-year-old man treated for lepromatous leprosy (LL) Hansen's disease presented with large joint pains for four months, for which he has been under treatment with oral prednisolone 60mg per day for four months from a quack. The patient gave a history of insidious onset of ill health for the past month with generalized weakness, anorexia, lassitude, and significant weight loss of 12 kg. He also complained of high-grade fever associated with chills and rigors, abdominal pain, abdominal distension, melaena for three days. Abdominal pain was acute, intermittent, stabbing in nature, localized to the epigastric region without any radiation, and was not related to food intake. There was a history of non-projectile, non-bilious vomiting for three days, six to eight episodes per day. There was no history of hematemesis. Rash was complained of over the thighs and abdomen, associated with mild pain, for three days. He is a known alcoholic for ten years, takes 90ml per day, and is a known smoker for ten years. He was diagnosed with LL Hansen's disease with bacteriological index (BI) of 4+ three years back at a government teaching hospital, for which he was given multi-bacillary multi-drug therapy for one year. Later on, he was alright for nearly two years when he developed the current illness.

Clinical examination revealed an ill-looking well built, ill-nourished individual. He was febrile with 100^{0}F, and pallor was present. Pulse and blood pressure were normal. There were about 15 tender erythematous nodules over both thighs, lower abdomen, flexor surface of the left forearm, both pinna (Fig. 1a, 1b). Painful joints were not swollen or erythematous. The card test was positive bilaterally. Trophic ulcers of 2x2 cm were seen over the right lateral foot and right heel. The left third toe got auto amputated. Numerous scaly

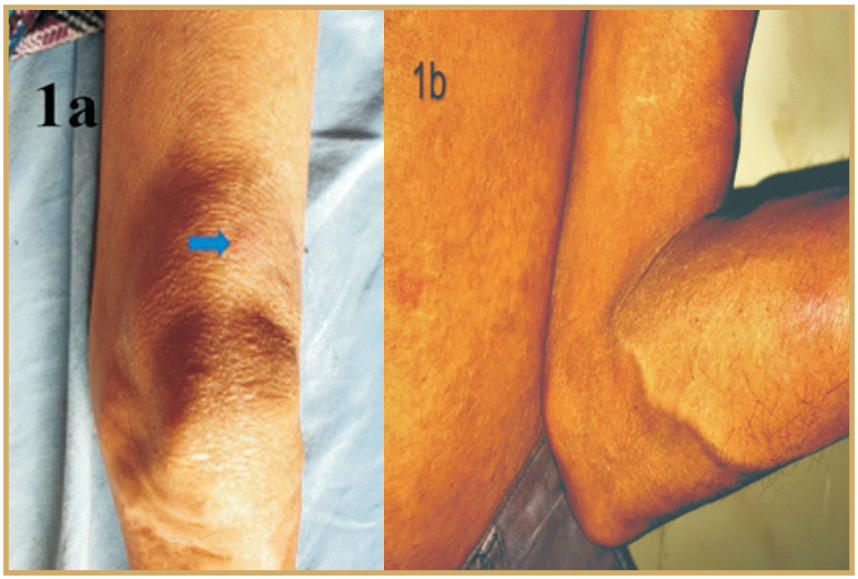

Fig. 1 : (a) An erythematous nodule over the left lower thigh. (b) Two erythematous nodules can be seen over the lower anterior abdominal wall and flexor left forearm.

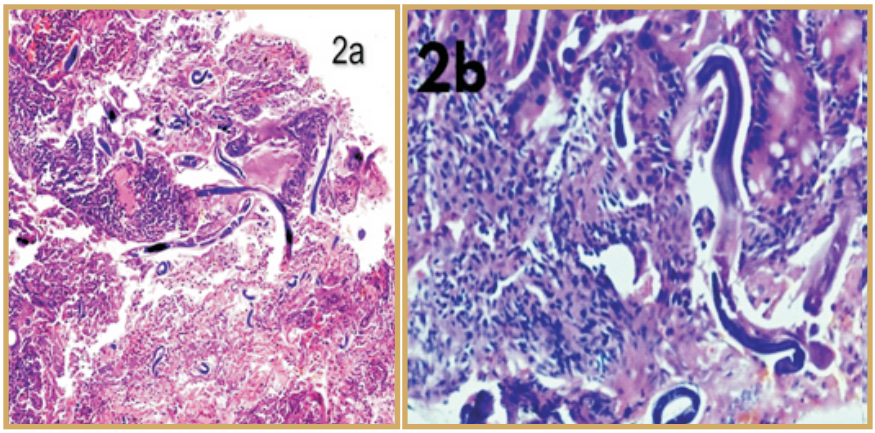

Fig. 2 : (a) Haematoxylin and Eosin (H&E) 4x and (b), H&E 40x. Histopathology of duodenal mucosa shows ova and larvae of Strongyloides.

annular lesions were present over the abdomen, thorax, both buttocks. Mild tenderness was present in the epigastrium, with abdominal distension and extension of the liver 2 cm below the right costal margin without any guarding or rigidity. Other systems were unremarkable.

Slit skin smear of the patient showed BI 3+. Peripheral smear showed normocytic normochromic blood picture with neutrophilic leukocytosis. Eosinophilia was absent. Chest radiography revealed bilateral pleural effusion. Enzyme-linked immune-sorbent assay test for human immunodeficiency virus-1 and -2, hepatitis B and C viruses were negative. The Monteaux test was negative. Rheumatoid factor was negative. Histopathology of the skin nodule confirmed erythema nodosum leprosum. Stool examination showed *Strongyloides* larvae and occult blood. Biopsy from duodenal mucosa revealed many *Strongyloides* larvae and ova within the mucosal crypts (Fig. 2a, 2b). The contrast-enhanced computerized tomography (CECT) abdomen revealed jejuno-jejunal intussusception (Fig. 3) and mesenteric lymphadenopathy. An upper gastrointestinal endoscopy showed pan gastritis with duodenitis and edematous duodenal mucosal folds. Based on these findings, the case was diagnosed as LL Hansen's disease with type 2 lepra reaction, bilateral ulnar nerve palsy, and acute abdomen due to jejuno-jejunal intussusception, caused by strongyloidiasis hyper infection, and or mesenteric lymphadenopathy, and tinea corporis. After

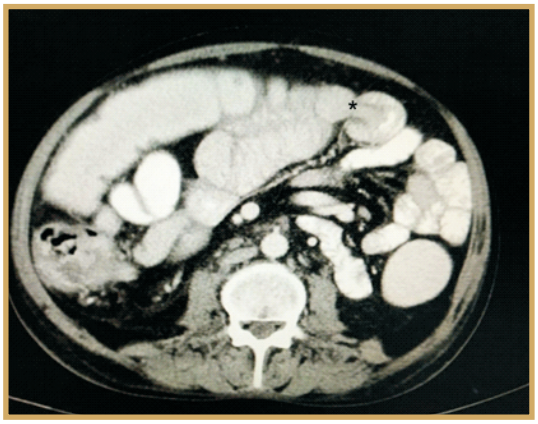

Fig. 3 : CECT abdomen shows the (*) jejuno-jejunal type of intussusception

admission, prednisolone was tapered and stopped. Albendazole 400mg twice daily for 14 days was given to treat Strongyloidiasis hyper infection and prevent disseminated infection. Intussusception was treated conservatively with supportive therapy. Abdominal symptoms gradually improved over two weeks. Prednisolone was tapered and stopped to avoid disseminated Strongyloidiasis infection.

Discussion

As the diagnosis of Strongyloidiasis is often overlooked, it has been uncommonly reported along with leprosy. Intestinal helminthic infestation can downgrade leprosy into more severe forms (Oktaria et al 2016). T-helper 2 (Th2) immune response in ENL is also predominant in helminthic infections. It has been suggested that soil-transmitted helminth infections were statistically significantly associated with a more severe type of leprosy and type II lepra reaction (Oktaria et al 2016). Lowered interferon-gamma and elevated Th2 cytokines interleukin - 4, 10 levels were found in lepromatous leprosy cases coinfected with soil-transmitted helminths (STH) (Oktaria et al 2016). STH may cause growth and dissemination of *Mycobacterium leprae* through the upregulation of Th2 cytokines or CD4+ CD25+ regulatory T-cells (Oktaria et al 2016).

Strongyloidiasis most commonly is an asymptomatic infection, but an alteration in host immune status can lead to fatal fulminant disease (Ramanathan & Nutman 2008). *S. stercoralis* has an auto-infective cycle in humans allowing it to persist indefinitely in the host. In immunosuppressed individuals, amplification of the normal life cycle of *S. stercoralis* can lead to accelerated autoinfection and hyper-infection. Occurrence of signs and symptoms due to enhanced larval migration through the organs involved in the auto-infective cycle (skin, lungs, gastrointestinal tract) helps diagnose *S. stercoralis* hyper-infection syndrome. On the other hand, disseminated infection refers to larvae in tissues other than these as well (Puthiakunnoon et al 2014).

Eosinophilia is often absent in severe infection due to steroid intake, as steroids lead to a decreased eosinophil count (Myint et al 2017). In our case, steroid intake has lead to reduced eosinophil count.

Definitive diagnosis is based on the detection of larvae in the stool or sputum samples. (WHO 2019). Positive yield with stool examination is not more than 40% (Sato et al 1995). This case shows larvae in the stool and duodenal mucosa, leading to a definitive diagnosis of Strongyloidiasis hyper-infection. Confirmation of diagnosis and follow-up is by serology as coprological diagnosis has low sensitivity due to irregular shedding of the worm (Oktaria et al 2016).

Strongyloidiasis infestation rate is higher in immunosuppressed people (Darlong 2016) and alcoholics (Teixeira et al 2016) due to the alteration of the immune system. Immuno-compromised status stimulates the transformation of rhabditiform larvae into the infective filariform larvae, leading to autoinfection. Challenge in treatment with steroids is the reactivation of asymptomatic infections (Bilodeau et al 2013). Ethanol intake might lead to an immune modulation or alteration in corticosteroid metabolism even in the absence of liver cirrhosis favoring *S. stercoralis* autoinfection. Another explanation is that chronic ethanol intake stimulates the hypothalamic-pituitary-adrenal axis to produce excessive endogenous cortisol levels, leading to a deficiency in Th2 protective response. Ethanol mimics the parasite hormone ecdysone, promoting the transformation of rhabditiform larvae to filariform larvae, leading to autoinfection (Teixeira et al 2016).

This patient is an alcoholic and took systemic steroids for four months for rheumatological manifestations (joint pains) of type two lepra reaction. Hence, chronic oral steroid intake (for four months), and alcohol could have been the reasons for strongyloidiasis hyper-infection in this case. Prior steroid use was a predisposing factor in 67% of severe or disseminated Strongyloidiasis. Even short courses (6–17 days) of corticosteroids have led to fatal hyper-infection (Bilodeau et al 2013), which may develop within four days after the onset of corticosteroid therapy and as late as several years, up to 20 years (Prabha & Chhabra 2018). One study estimated that dissemination occurs in 1.5–2.5% of all infected patients (Milder et al 1981). But the timely institution of albendazole in our case has possibly prevented dissemination of Strongyloidiasis infection.

Ascaris lumbricoides (Niang et al 2021) and mesenteric lymphadenopathy (Wang et al 2009) were described as two of the etiologies of adult intussusception. But *Strongyloides stercoralis* has not been mentioned till now as a cause of intussusception in adults with immunosuppression. Ascertaining whether *Strongyloides stercoralis* hyper-infection or mesenteric lymphadenopathy caused the intussusception, in this case, is difficult.

In Cambodia, the National Leprosy Program recommends administering empiric albendazole therapy before the initiation of corticosteroids. In Brazil, in some cases of leprosy, before initiating immunosuppressive treatment, therapy with anthelmintics is recommended to avoid hyper-infection. (Prabha & Chhabra 2018)

There is a need to know the incidence of strongyloidiasis in leprosy cases. If the incidence in an area is high, anthelminthic treatment may be initiated as prophylaxis in all leprosy cases to prevent more severe forms of leprosy, type 2 lepra reaction, and Strongyloidiasis hyper-infection.

Conclusion

Abdominal or lung symptoms in leprosy patients on chronic steroid therapy should raise the suspicion of Strongyloidiasis despite the absence of eosinophilia. Patients with Hansen's disease should be carefully screened with serial stool microscopic examination and specific serological antibodies for *S. stercoralis* and other intestinal parasitic infections, particularly in endemic areas. The positive result might necessitate pre-treatment with albendazole or ivermectin before initiating corticosteroid therapy to prevent severe forms of *S. stercoralis* infection.

In endemic areas, prophylactic administration of albendazole or ivermectin to all leprosy patients exposed to steroids or immunosuppressive agents is recommended.

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