Case Report

Strongyloides Hyperinfection Syndrome in Dermatology in-Patients: A Case Series

Chandra S. Sirka, Arpita N. Rout, Susama Patra[1], Hemanta K. Nayak[2]

Departments of Dermatology and Venereology, [1]Pathology, and [2]Gastroenterology, AIIMS, Bhubaneswar, Odisha, India

Abstract

Strongyloides stercoralis is a common intestinal pathogen, which often causes disseminated infection in patients on long term immunosuppressive therapy. The features of this syndrome range from subtle abdominal discomfort to fatal infection. Many of the immunologically mediated dermatological conditions need prolonged treatment with Immunosuppressants. Hence there is increased risk of this infestation. In dermatology in-patients, strongyloides stercoralis hyperinfection can be confused with a number of conditions, such as steroid-induced gastritis, corticosteroid withdrawal, electrolyte imbalance, lepra reactions, and erythrodermic enteropathy. In a country like India, where barefoot walking is still prevalent, this condition has been reported less often. We report a series of five patients who were diagnosed with strongyloides hyperinfection syndrome during their admission in the dermatology department during the last year. This case series aims at creating awareness among the dermatologists, so that, this, potentially fatal but easily preventable and treatable condition, can be managed properly.

Keywords: *Dermatology, immunosuppressives, ivermectin, round worm, strongyloides stercoralis*

Introduction

Strongyloides stercoralis can cause hyperinfection in immunosuppressed patients, which can be fatal, if left untreated.[1] Its nonspecific clinical features often lead to missed or delayed diagnosis. A large proportion of cases in dermatology in-patient need treatment with long-term systemic corticosteroids and other immunosuppressive agents, thus being at risk of developing such infections. The abdominal symptoms of hyperinfection with strongyloides can be easily confused with features of steroid-induced gastritis and nausea and vomiting associated with hypothalamopituitary axis suppression. We present a series of five cases diagnosed with features of strongyloides hyperinfection syndrome during last year in dermatology in-patients and treated successfully with ivermectin.

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Over last one year, total five cases, four of lepromatous leprosy with recurrent type 2 lepra reaction and one of chronic plaque psoriasis with recent aggravation as erythroderma, developed features like loss of appetite, vomiting, pedal swelling, head reeling, and abdominal discomfort. They had been on various immunosuppressants like oral prednisolone and methotrexate for variable periods, ranging from 2 months to 2 years. One patient had taken six months of indigenous ayurvedic preparations as treatment. All the patients were on normal Indian diet. The demographic details, primary dermatological diagnosis, doses and duration of immunosuppressive treatment, presenting symptoms, and investigation findings have been presented in Table 1.All patients were of average built, with normal weight and height. None of our cases had any cutaneous or pulmonary complaints suggestive of disseminated infection of strongyloidiosis.

All the cases, except case number 3, had pedal edema and anemia. Suspecting the possibility of suppression of hypothalamo-pituitary adrenal axis, the serum cortisol level was sent which was within normal limits in all of them. Stool sample on routine and microscopic examination did not show any abnormality in three cases, two cases showed multiple ova and larvae of strongyloides on routine

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Address for correspondence:
Dr. Chandra S Sirka, Department of Dermatology and Venereology, AIIMS, Bhubaneswar, Sijua, Patrapada, Odisha - 751 019, India.
E-mail: csirka2006@gmail.com

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Table 1: Enlisting the demographic details and the clinical presentation of the patients

Case number	Age	Sex	Diagnosis	Treatment	Duration	Symptoms	Investigations
1	24 years	F	Lepromatous leprosy erythema nodosum leprosum	MDTMB (A), prednisolone (40-60mg daily)	6 months	Loss of appetite, nausea, vomiting, abdominal discomfort, pedal swelling, weakness, head reeling	Stool R/M: WNL, Serum cortisol WNL, UGIE patchy erythema in the duodenal mucosa, endoscopic biopsy- strongyloides in various stages, hypoproteinemia, anemia
2	31 years	M	Lepromatous leprosy erythema nodosum leprosum	MDTMB (A), prednisolone (20-60mg)	24 months	Loss of appetite, nausea, vomiting, abdominal discomfort, pedal swelling	Stool R/M: 10-20 ova and larvae of strongyloides per high power field, Serum cortisol WNL, UGIE patchy erythema in the duodenal mucosa, endoscopic biopsy- strongyloides in various stages, hypoproteinemia, anemia
3	68yr	M	Lepromatous leprosy recurrent erythema nodosum leprosum	MDT MB (A), prednisolone (10-60mg)	8 months	Loss of appetite, vomiting weakness	Stool R/M: WNL, Serum cortisol WNL, UGIE patchy erythema in the duodenal mucosa, endoscopic biopsy- strongyloides in various stages
4	32 year	M	Lepromatous leprosy with recurrent erythema nodosum leprosum	MDTMB (A), prednisolone (10-40mg)	6months	Loose stool, pain abdomen, abdominal discomfort, loss of appetite, swelling of feet	Stool R/M: 4-6 ova and larvae per high power field, Serum cortisol WNL, UGIE patchy erythema in the antral mucosa, endoscopic biopsy- strongyloides in various stages, hypoproteinemia, anemia
5	57 years	M	Plaque psoriasis with erythroderma	Methotrexate 15-20mg once weekly, ayurvedic medications	2 months and 6 months respectively	Generalized edema, anemia, loss of appetite, vomiting	Stool R/M: WNL, Serum cortisol WNL, UGIE patchy erythema in the antral mucosa, endoscopic biopsy- strongyloides in various stages, hypoproteinemia, anemia

MDTMB (A): multidrug therapy multibacillary adult type, stool R/M: stool routine and microscopic examination, UGIE: upper gastrointestinal endoscopic examination, WNL: within normal limits

microscopy of stool sample. Diagnosis was confirmed in all cases by upper gastrointestinal endoscopy and biopsy. On upper gastro-intestinal endoscopy, all 5 patients showed patchy erythema with few erosions in the antrum of stomach and first and second parts of duodenum with esophagus showing normal mucosal study. Endoscopic biopsy showed body parts of Strongyloides embedded in duodenal and gastric mucosa [Figures 1 and 2]. Tests for HIV and hepatitis were negative in all the cases. Sputum examination and bronchoscopy was not done as there were no symptoms related to pulmonary system.

Antibody immunoglobulin level against strongyloides could not be done due to non-availability. Thus, with a final diagnosis of hyperinfection syndrome, the patients were treated with oral ivermectin 12mg on days 0, 1, 14, and 15. All the patients reported improvement in abdominal symptoms such as return of normal appetite, improvement in abdominal discomfort, nausea, and vomiting in 1–3 days. For case number 4 with loose stools, the stool frequency decreased and stool consistency improved gradually in 3 days.On subsequent follow-up visits, there was no clinical or microbiological evidence of hyperinfection.

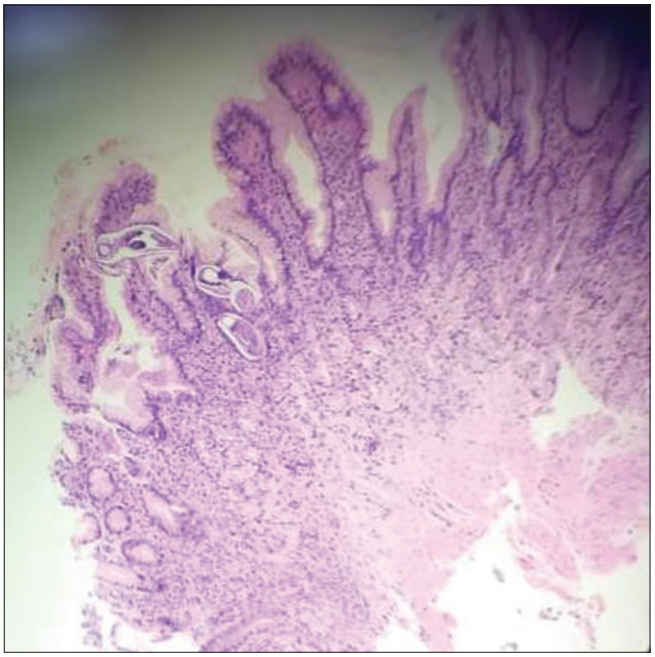

Figure 1: Scanner view showing intestinal mucosa with multiple larval forms and eggs of the organism *Strongyloides stercoralis* with inflammatory infiltrate. [H and E, 40x]

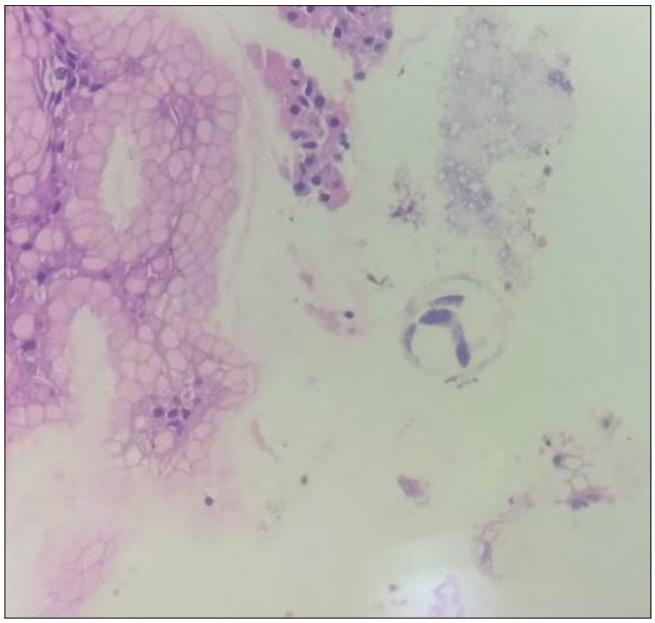

Figure 2: High power view shows multiple larval forms and eggs of *Strongyloides stercoralis* in the mucosa. [H and E, 400x]

Discussion

Strongyloides is a parasite that is prevalent in the tropical and subtropical regions of the world. Strongyloidiasis is caused by the female nematode *Strongyloides stercoralis*. In its classic life cycle, Strongyloides travels from the skin to the lungs and then to the gastrointestinal tract of its host. In hyperinfection syndrome, this classic life cycle is exaggerated (i.e. the parasite burden and turn around increase and accelerate) within the traditional reproductive route (the skin, gut, and lungs).[2] Disseminated disease is defined by the presence of parasites outside of the traditional life cycle (i.e. in organs other than the skin, gastrointestinal tract, or lungs), often involving the liver, brain, heart, and urinary tract.[2]

Patients acquire infection by walking barefoot in infested soils and can be infected for life because of the auto-infective cycle of the parasite. Corticosteroids have precipitated death in more than 60% of disseminated strongyloidiasis cases. There are few reports of hyperinfection syndrome in patients of leprosy with type 2 lepra reaction.[3-5]The male predominance in our series might be due to more outdoor activities. None of the patients had the history of taking any anti-helminthic treatment prior to initiation of prolonged therapy with steroid or other immunosuppressants as evident from the available prescriptions during presentation to our department.

The clinical manifestations of strongyloidiasis vary widely which include fever, abdominal pain, nausea, vomiting, diarrhea or constipation, gastrointestinal bleeding, cough, hemoptysis, dyspnea, and rarely, respiratory failure. Gram-negative sepsis is a dreaded complication that can prove fatal.[6]Dermatological manifestations include larva currens and purpura (the so-called "thumbprint sign" on the abdomen) and can be suggestive if present.

Intake of alternative medicines is common among Indians. These products are known to be adulterated with steroids.[7] Prolonged use of such products may lead to immunosupression and predispose such patients for hyperinfection syndrome.

In our patients, the diagnosis of *Strongyloides* hyperinfection was initially missed, as the nonspecific gastrointestinal complaints, anemia and hypoalbuminemia were attributed to the primary disease or the side effects of drugs. Anemia and hypoalbuminemia are often seen in leprosy, erythema nodosum leprosum, and cases of erythroderma. *Strongyloides* hyperinfection was an unexpected diagnosis in our cases, made on the incidental detection of the parasite on duodenal biopsy. Interestingly, anemia and hypoalbuminemia are common in strongyloidiasis as well, due to bleeding from intestinal erosions and ulcers and protein-losing enteropathy, respectively. Stool examination was negative in three cases, while the rest two cases showed plenty of ova and larval forms. The sensitivity of a single-stool examination is only 30%–50% which increases with repeated examination and using stool concentration techniques. Blood agar plate culture method and serological tests are more sensitive but have limited availability.

Treatment options include ivermectin, albendazole, mebendazole, and thiabendazole. Of these, ivermectin is preferred because of its better efficacy and tolerability.[6]

Strongyloides hyperinfection syndrome can be missed unless there is high degree of suspicion as the presenting complaints range from subtle to overt fatal infection. Dermatologists should be aware of this condition, so that early diagnosis and management could be done.

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Conflicts of interest

There are no conflicts of interest.

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