

# Intestinal Spirochetosis in a Patient with Celiac Disease

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## ABSTRACT

A young girl presented to us with recurrent diarrhea along with a history of 5 kg weight loss in one year. On examination, she appeared pale, while her laboratory reports showed a low hemoglobin, mean corpuscular volume (MCV) and serum albumin. Her erythrocyte sedimentation rate (ESR) was slightly raised with her iron profile suggestive of iron deficiency anemia. Viral markers, human immunodeficiency virus (HIV) serology along with thyroid profile were all unremarkable. There was no history of tuberculosis, and purified protein derivative (PPD) skin test was also negative. Computed tomography (CT) abdomen showed thickening of the terminal ileum with multiple enlarged lymph nodes. An esophagogastroduodenoscopy (EGD) along with colonoscopy was done. Multiple biopsies were taken, which were suggestive of sprue along with intestinal spirochetosis. Her tissue transglutaminase (TTG) was negative while deamidated gliadin peptide (DGP) was positive. She was kept on gluten-free diet and started on tablet metronidazole. This case shows that intestinal spirochetosis should be kept in mind in patients belonging to lower socio-economic status, who present with chronic diarrhea symptoms.

**Key Words:** *Intestinal spirochetosis, Celiac disease, Metronidazole, Chronic diarrhea.*

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## INTRODUCTION

Intestinal spirochetosis is a disease that occurs due to a wide variety of bacteria; and in humans, it is shown by the presence of spirochetes attached to the colorectal epithelium.<sup>1</sup>

Its prevalence is higher in areas where the living standards are poor.<sup>2</sup> A lower prevalence rate of around 2.6 to 16% is seen amongst humans living in the western countries.<sup>3</sup> Homosexual individuals and those with human immunodeficiency virus (HIV) carry a higher risk of being infected, along with higher chances of being symptomatic.<sup>2</sup> Children also have a higher rate of developing symptoms.<sup>1</sup>

Usually, intestinal spirochetes are discovered during routine colonoscopy as an incidental finding, while symptomatic disease presents as chronic diarrhea and abdominal pain.<sup>1</sup>

## CASE REPORT

A 16-year female presented to our outpatients' clinic with the complaints of recurrent episodes of diarrhea since the last two years. She had visited numerous physicians but her symptoms were still active at present. Her diarrhea was watery, yellowish in colour, small in amount, occurring 10-12 times a day, foul smelling,

associated with mucus and was difficult to flush away. It was associated with a diffuse crampy abdominal pain along with low grade fever. She had a history of unintentional weight loss of 5 kg during a span of one year. Her developmental history was unremarkable and had achieved all of her milestones on time. No history of tuberculosis was noted in the patient nor was there any prior history of tuberculosis exposure or any family history of malignancy.

On examination, the patient appeared pale with no visceromegaly appreciated. She was worked up for these complaints and her initial laboratory reports showed a low hemoglobin of 5.6 g/dL, mean corpuscular volume (MCV), 56 fL, a serum albumin, 2.3 mg/dl, while erythrocyte sedimentation rate (ESR) was raised 35 mm/hr. Rest of her laboratory reports were unremarkable. Her chest X-ray was normal, while serum iron profile was suggestive of iron deficiency anemia. Later on, her thyroid profile was sent, which was within the normal limits as well (TSH, 1.9 mIU/L). Her viral markers, and human immuno-deficiency virus (HIV) serology were negative. Stool detailed report, culture and sensitivity report also tested negative.

Purified protein derivative (PPD) skin test was done and it showed an induration of less than 10 mm at 72 hours.

Later on, CT abdomen was done, which showed mild thickening in the terminal ileum along with multiple lymph nodes, the largest one in the ileocecal region, measuring 1.6x0.8 cm.

She underwent an upper GI endoscopy along with colonoscopy, both of which were normal; and multiple biopsies were taken (from the terminal ileum, cecum, ascending colon, transverse colon, descending colon, rectosigmoid and duodenum).

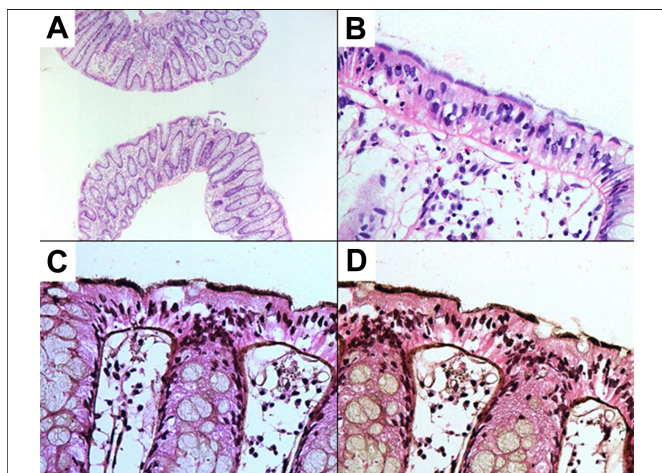
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**Figure 1:** Intestinal spirochetosis. (A) Low-power view showing overall preserved mucosal architecture and minimal inflammation. Organisms are not apparent. (H&E,  $\times 50$ ). (B) High-power view showing "blue fringe" on the luminal surface of epithelial cells. (H&E,  $\times 400$ ). (C) Silver staining highlighting the silver-positive organisms. (JMS,  $\times 400$ ). (D) Another area of the biopsy showing silver-positive organisms on the luminal surface of the epithelial cells. (JMS,  $\times 400$ ).

The biopsies of the terminal ileum and duodenum showed sprue, Marsh class 3A, while the biopsy of the cecum revealed intestinal spirochetosis. While biopsies of the rectosigmoid, ascending colon, descending colon revealed mild, chronic, non-specific inflammation. No evidence of tuberculosis was noted in biopsies.

Based upon her biopsy report, her tissue transglutaminase (TTG), and deamidated gliadin peptide (DGP) were sent and DGP came out positive.

She was diagnosed as a case of celiac disease along with intestinal spirochetosis; and after consultation with a dietician, she was kept on gluten-free diet and started on tablet metronidazole. Later on, after a span of one month, her symptoms improved and she was planned for a follow-up endoscopy.

## DISCUSSION

In humans, intestinal spirochetosis is reported to cause diarrhea along with rectal bleeding,<sup>4</sup> and is usually not seen in children. Diagnosis is based upon a high degree of clinical suspicion amongst those presenting with abdominal pain, hematochezia, symptoms of chronic diarrhea while having a normal endoscopic examination. Hence, multiple colonic biopsies are recommended for its diagnosis.<sup>5</sup>

Helbling *et al.* showed intestinal spirochetosis in a child mimicking inflammatory bowel disease.<sup>5</sup> King *et al.* showed it to be presenting as rectal bleed in a young child.<sup>6</sup>

Histologically, it appears as a false brush border on the colonic mucosa epithelium.<sup>7</sup>

Identification of spirochetes is done *via* the use of the hematoxylin and eosin (H and E stain) (Figure 1), while the most sensitive test for diagnosing it is polymerase chain reaction (PCR).<sup>7</sup> Since, it is hardly seen in children, proper treatment protocols are lacking. A review of the medical literature suggests the best treatment protocol as macrolide antibiotics with or without metronidazole.<sup>8</sup>

Invasion of the spirochetes beyond the epithelial surface leads to diarrhea, which resolves on treatment with metronidazole.<sup>2</sup>

It has also been noted that intestinal spirochetes may result in colorectal disease in the pediatric population; hence, early identification and treatment can be fruitful.<sup>9</sup>

In summary, intestinal spirochetosis has rarely been reported in the pediatric population in the medical literature. To the best of the authors' knowledge, this represents the first case reported so far, of a child having intestinal spirochetosis along with celiac disease.

We suggest that intestinal spirochetosis should be kept in mind in those patients having chronic diarrhea. Whether patients with celiac disease are more prone to infection with spirochetosis is still not known.

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