

Intestinal Spirochetosis: Symptomatic Patients in the Presence of No Risk Factors and Improvement with Metronidazole Therapy

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Human intestinal spirochetosis, which was first described by Harland and Lee in 1967, is an infestation of spirochetes usually present on the surface of the large intestinal mucosa [1]. Causative spirochetes are reported to be either *Brachyspira aalborgi* or *Brachyspira pilosicoli*. The two species may be zoonotic because they have been isolated from the feces of non-human primates and other animals. Spirochetes in humans and other species presumably result from fecal-oral contamination [2]. The prevalence of spirochetosis in humans varies from 2.5 to 16% in Western countries [3]. The prevalence in homosexual and immune-compromised patients, based on stool culture and biopsy findings, is as high as 50%. The colonization rate in human colon is variable and is influenced by many factors, including immune function, sexual practices, diet, sanitation, and community structure [3].

In humans, the pathological and clinical significance of these organisms is far less clear and controversial, although there have been reported cases associated with rectal bleeding and diarrhea. It still remains controversial that whether these spirochetes are potential pathogens or non-pathogenic commensals as a part of

Figure 2: The architecturally normal appearing mucosa with a blue hematoxyphilic line seen along the luminal border for which PAS, Giemsa and Warthin-Starry stains were positive

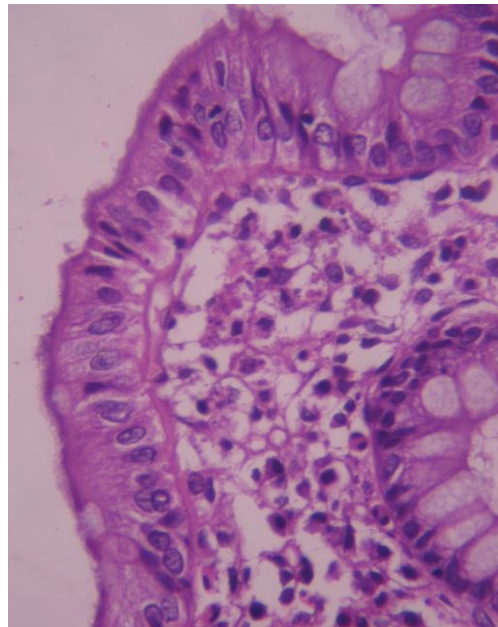


Figure 1: Normal sigmoidoscopic view



normal flora because most cases are asymptomatic [2]. Thus, when the diagnosis of human intestinal spirochetosis is confirmed, the clinicians usually adopt a 'wait and see' policy. As to possible correlation to other intestinal diseases, human intestinal spirochetosis is frequently associated with various intestinal diseases, such as carcinoma, adenomatous polyp, metaplastic polyp, and ulcerative colitis, and that colonic carcinoma was the most frequent because chronic stasis of intestinal content favors the infestation of the spirochetes [3].

Spirochetes can be seen easily by light microscopy as a fuzzy blue, 3-7 micron thick, hematoxyphilic line on the luminal border of the large intestine, which may be confused with glycocalyx and distinct larger organisms [4]. The histological appearance of this organism is that of a false brush border on the colonic mucosa, whi-

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ch represents a layer of spirochetes, especially *Brachyspira aalborgi* and *Brachyspira pilosicoli* (formerly *Serpulina pilosicoli*). Spirochetes can be easily demonstrated using silver stains, such as Warthin-Starry. They also stain, although less intensely, with Giemsa, periodic acid-Schiff (PAS), and Alcian blue at pH 2.5. The inflammatory response is usually minimal or absent. Immunohistochemistry using polyclonal spirochete antibodies may facilitate the histological diagnosis, but generally is not very helpful. Electron microscopy and molecular studies can also be used not only to identify the spirochetes, but also to help determine the species of the spirochete. Spirochetes are difficult to grow on culture media owing to their fastidious nature. They grow poorly or not at all in the absence of serum supplements. Due to this limitation, there is no published data characterizing the organisms. Spirochetes respond very well to metronidazole 400 mg TID for 10-14 days course in most cases, but not always [5].

CLINICAL CASES

We report two patients as among the first few cases of intestinal spirochetosis from Islamabad, Pakistan who were incidentally diagnosed during routine evaluation for chronic diarrhea.

The first patient was a 45 years old male, junior scientific laboratory technician, normotensive, normoglycemic presented with a one-year history of on and off diarrhea and abdominal cramping. He had watery diarrhea of frequency 3 to 4 times per day without any blood or mucus in it at present. In the past medical history, he suffered from 3-4 episodes of diarrhea containing blood in last one year but was not investigated for etiology at that time. While the second patient was a 37 years old male, senior scientific laboratory technician, normotensive, normoglycemic presented with a 5-month history of on and off diarrhea and abdominal cramping. He had watery diarrhea of frequency 3 to 5 times per day without any blood or mucus in it with no significant past medical or surgical history. Both the patients had no history of nausea, vomiting, fever or significant weight loss with unremarkable remaining systemic enquiry. These patients had no history of smoking, alcoholism, drug abuse or homosexuality and also denied any zoonotic contact. Clinical examination was unremarkable.

Both patients underwent a complete work-up, including detailed history and physical examina-

tion complete blood count, erythrocyte sedimentation rate, urine analysis, stool studies for ova and parasites, *Cryptosporidium*, *Clostridium difficile* toxin, testing for human immunodeficiency virus, Hepatitis B, C and sexually transmitted diseases as well as blood, urine and stool cultures, all of which were negative.

These patients also underwent endoscopic examination of colon, which showed normal appearing mucosa in both cases (Figure 1). However, histopathological examination of two random biopsies taken from each patient showed architecturally normal appearing mucosa with minimal focal signs of inflammation. A brush border like blue hematoxyphilic line was seen along the luminal border for which PAS, Giemsa and Warthin-Starry stains were positive in both cases (Figure 2). In conclusion, sigmoid colon biopsy findings revealed the diagnosis of intestinal spirochetosis in both cases.

Both patients were treated with oral metronidazole 400 mg three times daily for 2 weeks with resolution of their symptoms within the first few days. At 6 weeks, we offered repeat endoscopic examination of colon to document histopathological response but both patients refused.

We report these patients because these were among the first few cases from Pakistan that were found incidentally on routine evaluation for chronic diarrhea within 3 months. These patients were free of common risk factors such as poor hygiene, zoonotic contact, alcoholism, drug abuse, homosexuality, immunodeficiency or concomitant intestinal disease. Both patients improved clinically with 2-week course of metronidazole. This suggests the pathogenic potential of intestinal spirochetes and response to treatment with metronidazole. We suggest further detailed studies regarding intestinal spirochetosis to formulate management guidelines.

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