



Intestinal Spirochetosis: An Interesting Cause of Chronic Diarrhea

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Abstract

Forty-six-year male from India presented with 2-year history of small volume non bloody diarrhea with mild abdominal pain. He had no history of fever or constitutional symptoms and only minimal weight loss. His blood evaluation showed mild iron deficiency anemia with normal serum albumin and renal profile. Routine stool examination showed no blood, pus cells, ova or parasites. Celiac serology was negative. Contrast enhanced CT (Computed Tomography) abdomen showed subcentimetric mesenteric lymph nodes, bowel was normal in size and thickness. Upper GI endoscopy was normal with normal duodenal biopsy; ileocolonoscopy showed few aphthous ulcers in terminal ileum with mild nodularity. Ileal biopsies showed bluish filamentous structure on surface epithelium forming a thick false brush border appearance, suggestive of intestinal spirochetosis. Rest of ileal architecture was well preserved.

Patient was commenced on cyclical antibiotics including Nitazoxanide, Metronidazole, doxycycline and Rifaximin. Patient on follow up showed improvement in symptoms and weight gain.

Intestinal spirochetosis is commonly transmitted to human from infected animals. Our patient was butcher by occupation. Our case is an interesting presentation of Intestinal Spirochetosis in form of chronic diarrhea.

Keywords: Spirochetosis; Chronic diarrhea; False brush border

Introduction

Spirochete induced diarrhea is common in veterinary medicine, which is observed in swine, poultry, dogs, cats and non-human primates [1]. Humans are usually affected due to close proximity and is common in animal handlers like poultry workers, butchers, etc. Other common association is seen amongst homosexual men and immunocompromised individuals. Most commonly detected as an incidental finding during screening colonoscopy, co-infection [2] with intestinal pathogens like *Shigella*, *Enterobius vermicularis*, and *Helicobacter pylori* makes intestinal spirochetosis of questionable clinical significance. Colonoscopic appearance seldom offers any clue to diagnosis. Histologic examination is the key to diagnosis which presents as “false brush border” on Hematoxylin-Eosin staining [3]. Electron microscopy is the gold standard diagnostic test, but not often available in clinical setting. Metronidazole is the usual choice of drug but various antibiotics like Clindamycin, macrolides have been reported of utility [4].

Case Presentation

A forty-six-year male presented with a two-year history of non-bloody diarrhea. His stool frequency was 4 to 5 per day small quantity, on most days with no nocturnal frequency. He complained of mild lower abdominal pain which would subside post defecation. He had no history of fever, weight loss of 1 to 3 kilogram over the two-year duration. He was butcher by occupation, denied any homosexual behavior or use of habitual drugs or medications. He was non-diabetic, not on any immunosuppressive medicines and denied significant travel history, alcohol intake or drug abuse. Clinical examination showed normal vital parameters, no clubbing or lymphadenopathy or edema.

Routine blood investigations revealed hemoglobin of 12 gm/dl, normal leucocyte count (7500/cmm) and normal iron profile (Iron 120 mcg/dl, TIBC 300 mcg/Dl, Transferrin saturation 30%) His serum protein was 7 gm/deciliter and Albumin of 4 gm/deciliter. Celiac serology was negative. Stool examination showed no red cells, pus cells or ova and parasites.

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Figure 1: Colonic mucosa was essentially normal.

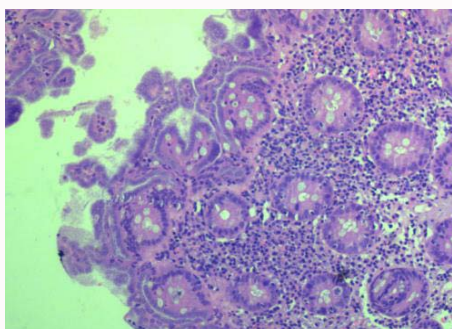


Figure 2: Maintained architecture.

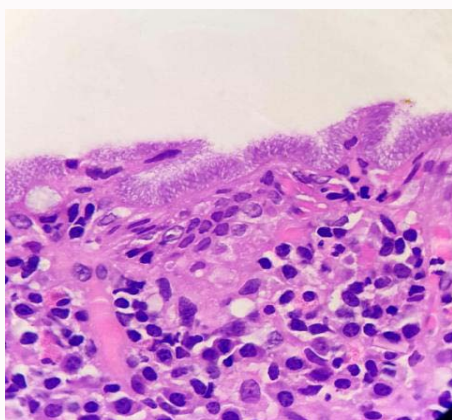


Figure 3: "False brush border" like appearance.

A contrast enhanced Computed Tomography (CT) abdomen showed normal bowel wall thickness and revealed small mesenteric lymphadenopathy.

Patient was subjected to Gastroscopy and Ileocolonoscopy as evaluation of chronic diarrhea and was found to have normal duodenal mucosa. Ileocolonoscopy revealed few aphthous ulcers in terminal ileum with mild nodular mucosa (Figure 1).

Colonic mucosa was essentially normal. Multiple biopsies were obtained from duodenum, ileum and colonic mucosa. Duodenal biopsy showed maintained architecture, no features of Celiac disease. Ileal biopsies (Figure 2) showed maintained architecture. There was mild increase in acute inflammatory cells, namely polymorphs and eosinophils. Few lymphocytes were noted within muscularis mucosa and submucosa. No granuloma or cryptitis, crypt abscesses were

found. Hematoxylin - Eosin staining showed bluish filamentous structure on surface epithelium forming a thick "false brush border" (Figure 3) like appearance, suggestive of Intestinal spirochetosis. Patient responded to cyclical therapy consisting of metronidazole, nitazoxanide and rifaximin which resulted in complete clinical response over 6 months.

Discussion

Intestinal Spirochetosis (IS) is a term coined by Harland and Lee dating back to 1967, where he recognized adherence of spirochetes to colonic mucosa in histopathologic examination and confirmed on electron microscopy.

Despite improved detection, whether IS represents a disease entity in humans or just incidental intestinal colonizer is debatable. IS is more prevalent in developing countries and regions as evident in many recent reports from Australia where it was identified amongst aboriginal Australians much more than developed regions within the continent [5]. Various other studies showed similar trend with more prevalence (1.1% to 5%) amongst rural and suburban areas compared to urban population [6-8]. Higher rates of intestinal colonization of IS is seen in homosexual men and Human Immunodeficiency Virus (HIV) infected individuals. Homosexuality was thought to be more responsible rather than immunodeficiency per se [9].

Spirochetes are divided into three polygenetic groups, namely *Spirochaetaceae* (*Borrelia*, *Spirochaeta*, *Spironema* and *Treponema*), *Leptospiraceae* (*Leptonema*, *Leptospira*) and *Brachyspiraceae* (*Brachyspira*). Intestinal spirochetosis is most commonly associated with caused by two members of the *Brachyspiraceae* family, most commonly associated with human IS are *Brachyspira aalborgi* and *Brachyspira pilosicoli* [10].

As far as clinical presentation is concerned, symptomatic IS are accompanied by chronic diarrhea with vague abdominal pain with or without signs of malabsorption. Few severe cases of invasive infection are identified in literature; however, most IS are incidentally diagnosed on histologic examination. Co infection due to *Helicobacter Pylori*, *Shigella flexneri* or *Enterobius vermicularis* are seen in many cases raising question about clinical importance of IS.

Endoscopic appearance seldom offers any clue to diagnosis, and histologic examination of colonic mucosal biopsy mostly gives the diagnosis of IS. The diagnosis of IS is traditionally based on the histological appearance of a diffuse blue fringe (seen in hematoxylin-eosin stain), which is approximately 3 μ m to 6 μ m thick, along the border of the epithelial lining. This finding is referred to as the "false brush border" [4]. Electron microscopy is the gold standard test for confirmatory diagnosis available at few institutes. Newer techniques like PCR based tests (targeting 16sRNA, NADH oxidize, 23rDNA gene) and Fluorescent in Situ Hybridization [FISH] are done in research setting and yet to be available in clinical practice [11,12].

Treatment response is variable as many cases are asymptomatic and few are accompanied by superimposed bacterial infection. Available data suggests good response to a ten-day therapy with metronidazole [13]. Other alternatives used are clindamycin, macrolides are other alternatives.

In our case, patient presented with chronic diarrhea and mild abdominal pain. His colonoscopy showed few aphthous ulcers in terminal ileum and normal colonic mucosa. Histopathology was diagnostic and beautifully demonstrated the classical "False brush

border” sign. Patient responded to cyclical therapy consisting of metronidazole, nitazoxanide and rifaximin which resulted in complete clinical response over 6 months. Our case is unique in the sense it was evident in an immunocompetent patient with chronic diarrhea and without any severe constitutional symptoms.

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