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Intestinal Spirochetosis, A Rare Cause of Abdominal Pain in A Polymorbid Immunocompetent Patient: A Case Report

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ABSTRACT

Intestinal spirochetosis is a rare infection of the gastrointestinal tract caused by the colonization of in colonic mucosae by spirochetae. We report a case of a 78-year-old immunocompetent polymorbid woman who was referred to the gastroenterology clinic for a diagnostic colonoscopy because of unclear chronic abdominal pain. The diagnostic colonoscopy revealed diffuse edematous mucosa in the entire colon and sigma with presence of Spirochetes in the biopsy specimens. The patient was treated with metronidazole 500mg four times a day for ten days. After treatment, we observed a complete remission of symptoms. Chronic pain in these patients is often wrongly considered to be related to previous surgery or polymorbidity. The endoscopic finding of edematous mucosae was non-specific and could not lead to the diagnosis without performing biopsies for histological investigation. Our case underlines the importance of performing diagnostic colonoscopy in immunocompetent polymorbid patients with chronic abdominal pain.

Keywords: *Intestinal Spirochetosis, Diagnostic Colonoscopy, Chronic Abdominal Pain, Spirochetae*

Introduction

Intestinal spirochetosis is a rare infection of the gastrointestinal tract caused by the colonization of in colonic mucosae by spirochetae. Immunocompetent patients are mostly asymptomatic, but in certain cases, abdominal pain, diarrhea or bleeding are reported. Because of unspecific clinical presentation and normal endoscopic findings, intestinal spirochetosis is often missed and the cause of symptoms remains unexplained. We report a case of chronic abdominal pain caused by colonic spirochetosis in an immunocompetent patient with a significant medical past history and polymorbidity.

Case Description/Methods

A 78-year-old immunocompetent woman was referred to the gastroenterology clinic for a diagnostic colonoscopy because of unclear chronic abdominal pain. The patient had been seen in the emergency department eight months before for a burning sensation in the stomach, retrosternal pain and weight loss. Her past medical history included surgical thoracic spine stabilization eleven months before the current presentation and post-surgery complications with removal of a retroperitoneal hematoma ten days later. Moreover, she was known for osteoporosis under treatment with denosumab, chronic kidney failure and intraductal papillary mucinous neoplasm (IPMN) in the uncinate process. Laboratory findings in the emergency department showed slightly elevated C-reactive protein (CRP; 10 mg/L) with normal white cells count, hypochromic microcytic anemia: hemoglobin (Hb) 103 g/L, mean corpuscular volume (MCV) 78fL, mean corpuscular hemoglobin (MCH) 24 pg and elevated D-dimers (2868 µg/L). A cardiologic work-up and a computed tomography pulmonary angiogram (CTPA) were normal. A gastroscopy and upper endosonography were performed in nineteen months bevor current presentation and showed no pathological findings except the known pancreatic IPMN which was stable in dimension. The symptoms were attributed to gastroesophageal reflux disease (GERD) and concomitant psychological burden and the patient was discharged with high dose pantoprazole and mirtazapin. At the gastroenterology outpatient clinic, the patient presented in good general condition with an unremarkable physical exam. She reported chronic diffuse abdominal pain, which was not meal-related, and denied B symptoms, dyspepsia, diarrhea, melena, and hematochezia. The family history was negative for colorectal cancer. The diagnostic colonoscopy revealed diffuse edematous mucosa in the entire colon and sigma as well as colonic diverticulosis (Fig. 1).

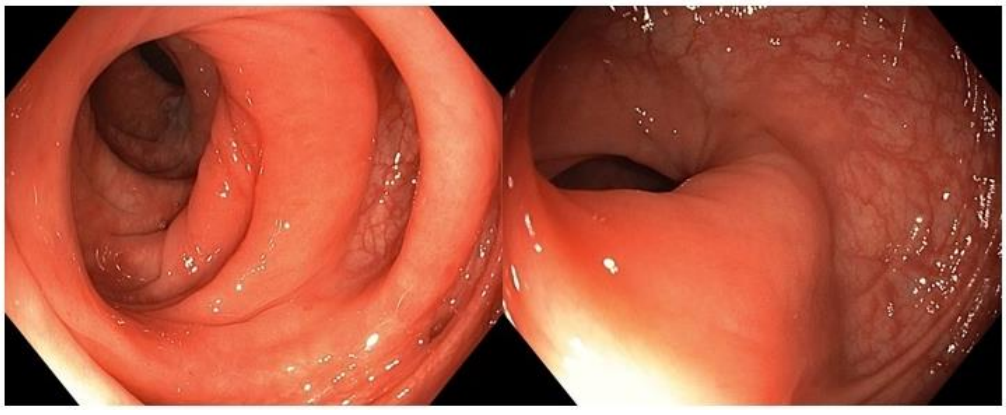

Figure 1: Edematous mucosal folds with partial loss of vascular pattern in the left colon (A) and in the sigmoid colon (B)

The biopsies taken from the ascending-transversum-descending colon and sigma showed a typical luminal blue fringe with unremarkable surface epithelium. Periodic acid-Schiff reaction (PAS) confirmed the presence of Spirochetes (Fig. 2 and Fig. 3).

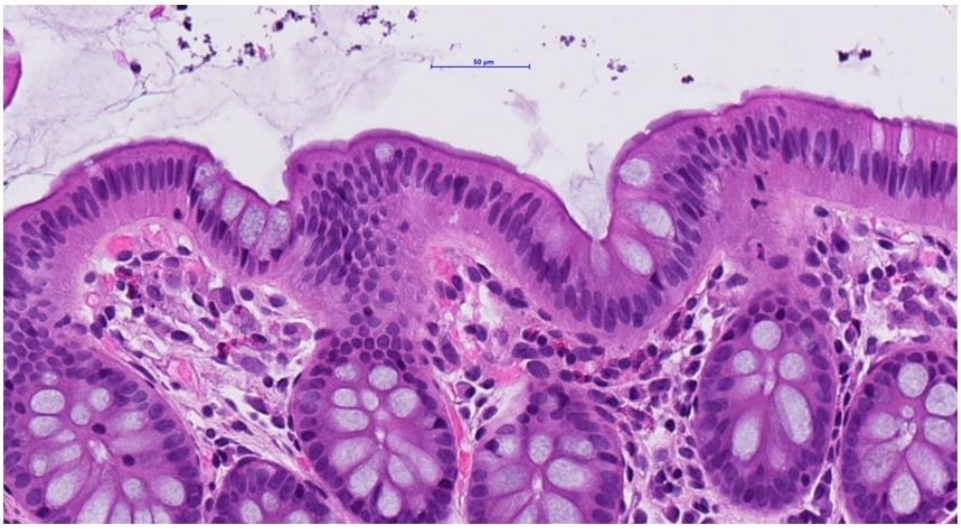

Figure 2: Colonic mucosa with a luminal blue fringe on the unremarkable surface epithelium with presence of Spirochetes lining the mucosa forming a false brush border (HE staining)

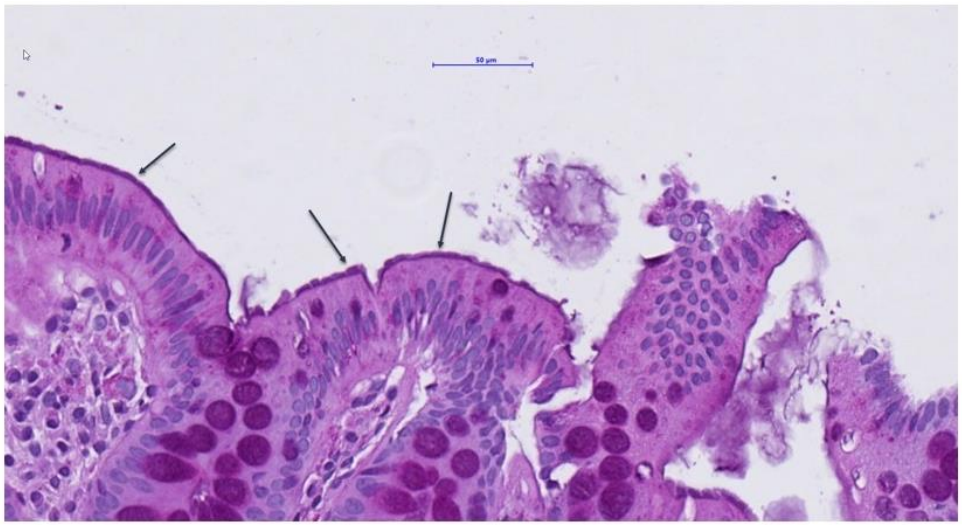

Figure 3: Spirochetosis on the surface of colonic epithelium, highlighted as a purple hem (arrows; PAS staining)

Due to this finding, the patient was treated with metronidazole 500mg four times a day for ten days. After treatment, we observed a complete remission of symptoms.

Discussion

Intestinal spirochaetosis (IS) is rare infection of the gastrointestinal tract caused by the overgrowth of gram-negative bacteria in colonic mucosae. Most of the time, the course of infection is asymptomatic but in some cases it can be associated with various abdominal symptoms including pain, diarrhea and rectal bleeding, especially in immunocompromised and homosexual patients (Amat *et al*., 2004; Harland and Lee, 1967; Paul *et al*., 2004; Körner and Gebbers, 2003; McMillan and Lee, 1981). Many organism within the Spirochaetes phylum are pathogenic: Leptospira species, which causes

leptospirosis, Borrelia burgdoferi, Borrelia garinii, and Borrelia afzelii, which cause Lyme disease, Borrelia recurrentis which causes relapsing fever, Treponema pallidum subspecies which cause treponematoses such as syphilis and yaws. Finally, Brachyspira pilosicoli, Brachyspira aalborgi and Serpulina pilosicoli are most commonly associated with human IS (Wolgemuth, 2015). The prevalence of spirochetosis is based on biopsy findings and varies from 2 to 7% in western countries, 1-34% in less developed countries and up to 54% in men who have sex with men and patients with human immunodeficiency viruses (HIV), suggesting sexual transmission in some cases (Körner and Gebbers, 2003; McMillan and Lee, 1981).

The endoscopic appearance of the colon normally does not contribute to the diagnosis and varies from normal appearing mucosa to erythematous mucosa and in some cases is also found incidentally in association with adenomatous or hyperplastic polyps, diverticular disease and inflammatory bowel disease (Alsaigh and Fogt, 2002). Histologically, intestinal spirochetosis is characterized by a distinctive basophilic, fringe-like, end-on-end attachment of densely packed filamentous spirochetes along the border of the intercryptal epithelial layer, known as "false brush border" (Harland and Lee, 1967). This finding can be seen in the hematoxylin-eosin (H&E) stain; however, Warthin-Starry or PAS are stains usually used for further clarification (Tsinganou and Gebbers, 2010). The most appropriate means of identification are polymerase chain reaction-based techniques, which either target 16S ribosomal ribonucleic acid (rRNA), nicotinamide adenine dinucleotide hydride-oxidase (NADH-oxidase) or the 23-ribosomal deoxyribonucleic acid (23rDNA) gene. However, these tests are costly, which is particularly important in developing countries (Esteve *et al*., 2006; Schmiedel *et al*., 2009). Even in symptomatic patients, the organism have been described to be typically non-invasive mainly attached to the luminal border of colonocytes mostly without actually penetrating the membrane. Luminal colonization causes epithelium changes such as blunting and loss of the microvilli (Tsinganou and Gebbers, 2010). Invasive intestinal spirochetosis is characterized by cell-membrane destruction, in this case spirochetes are found in the intercellular spaces, within the surface epithelial cytoplasm. The amount of cell destruction is usually proportional to the degree of invasion, with more diarrhea typically seen in those with a greater degree of microvillous destruction and a higher burden of spirochete attachment. The mechanism of diarrhea is believed to be a result of decreased resorptive areas of the damaged brush border (Tsinganou and Gebbers, 2010).

Our case underlines the importance of performing diagnostic colonoscopy in immunocompetent polymorbid patients with chronic abdominal pain. Chronic pain in these patients is often wrongly considered to be related to previous surgery or polymorbidity. The endoscopic finding of edematous mucosae was non-specific and could not lead to the diagnosis without performing biopsies for

histological investigation. Histologically the classic presentation of a non-invasive intestinal spirochetosis with intact villi was concluded to explain the diarrhea and/or bleeding. Most important, there were no other signs of acute or chronic inflammatory bowel disease. The treatment of intestinal spirochetosis is based on the clinical presentation, severity of symptoms, and immune status. In our case, we prescribed a course of antibiotic therapy with metronidazole 500 mg qid for ten days. After this therapy, the patient reported a complete remission of symptoms, suggesting that her symptoms were indeed related to the diagnosed IS. Almost one year later, the patient reports no recurrence of abdominal pain and/or other gastrointestinal symptoms. Because of life threatening complications in immunocompromised patients or disseminated infection, an infectious disease specialist may be advised in these cases

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