

Chronic Diarrhea Due to Intestinal Spirochaetosis in an HIV-Positive Patient: Case Report and Literature Review

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Abstract

Intestinal spirochaetosis refers to the colonization of the colorectal epithelium by anaerobic spirochetes of the genus *Brachyspira*. It is more frequently identified in developing countries or when sanitation conditions are suboptimal. It can affect anyone, although it is more common in gay men, particularly those infected with the human immunodeficiency virus (HIV). Its most common symptom is chronic watery diarrhea. A colonoscopy may be expected. We present the clinical case of a young male patient with chronic diarrhea of several years of evolution resolved by specific treatment. In addition, a review of the literature on this disease is provided.

Keywords

Brachyspira, diarrhea, colonoscopy, *Spirochaetales*.

INTRODUCTION

Chronic diarrhea (greater than or equal to 28 days) is a frequent reason for consultation in primary and specialized medical care and can be present in up to 28% of patients with human immunodeficiency virus (HIV). A study with this population group found that up to 40% had at least one diarrheal episode in the month before being interviewed, and it was severe (> 6 stools/day) in 3% of them⁽¹⁾. Although the etiology of this diarrheal disease can be multifactorial, from infectious causes, drugs, and HIV enteropathies to malignancy, it is not identified in many cases after a meticulous search for its etiology. An unusual case of chronic diarrhea is reported in an adult patient infected with HIV who was documented to have the colonic mucosa colonized by spirochaetes.

CLINICAL CASE

We present the case of a 31-year-old male patient, homosexual, diagnosed with HIV infection 12 years ago. He consulted due to a picture of over 5 years of intermittent diarrhea lasting several months, each associated with rectal bleeding. For this reason, he has undergone multiple colonoscopies, all identifying moderate inflammatory changes and a rectal ulcer. The histopathological diagnoses of the serial biopsies were nonspecific, and, as relevant data, one reported mild acute proctocolitis in the rectosigmoid. In the ulcer, nonspecific chronic inflammation and severe acute inflammation with granulation tissue were documented. In a biopsy of the anal canal, cytopathic changes suggestive of human papillomavirus (HPV) infection were noted. Among the multiple empirical treatments, he recei-

ved mesalazine without improvement and treatment for syphilis due to a non-treponemal serological test, which was reported positive with 16 dilutions without improvement in his symptoms.

In our service, a new ileocolonoscopy requested on an outpatient basis was performed in July 2021, in which severe inflammation and friability of the mucosa with fibrin-covered ulcers were observed in the distal rectum and anal canal. The rest of the mucosa had a normal appearance. The ileum, colon, and rectum samples were taken for histopathological study.

He was evaluated by outpatient consultation in our service in November 2021 due to persistent symptoms, so inflammatory bowel disease was suspected. By then, he had an undetectable viral load, CD4 of 699 cells/mm³, stool analysis without abnormalities, and negative stool culture. There were no abnormal findings on physical examination. The histopathological study report identified luminal filamentous microorganisms compatible with intestinal spirochaetosis in the samples of ileal mucosa, cecum, and ascending and transverse colon. Granulation tissue and reactive epithelial changes were identified in the rectal mucosa samples without granulomas or viral cytopathic effect.

Management involved metronidazole 500 mg orally every 8 hours for ten days, with which he reported complete improvement in diarrheal stools and rectal bleeding. A control colonoscopy was not performed due to the resolution of the clinical condition.

DISCUSSION

The term *intestinal spirochaetosis* was coined in 1967 by Harland and Lee to describe the colonization of the epithelial cells of the colon and cecal appendix by anaerobic spirochaetes, which can be recognized in a histopathological study or electron microscopy^(2,3,13); observing them is essential for the diagnosis. The mucosa of the terminal ileum may also be colonized⁽³⁾. This occurs due to two different species: *Brachyspira aalborgi* (considered a commensal pathogen) and *Brachyspira pilosicoli* (more commonly in an asymptomatic form, but sometimes as an opportunistic pathogen)^(3,4,5).

The frequency of intestinal spirochaetosis in the general population is low. It has been regarded as a disease associated with states of immunosuppression, but it can also occur in immunocompetent patients⁽³⁾. There are higher-risk groups in which it occurs more frequently, such as men who have sex with men, especially those with HIV. However, it is also identified in HIV-negative people in fewer cases. It is believed that one route of infection is sexual, probably due to oroanal (odds ratio [OR]: 3.45) (4.5) and anoreceptive sexual practices, and another is

orofecal due to the ingestion of water contaminated with animal feces. It is more prevalent in developing countries, probably due to the ingestion of untreated water^(5,7). In Western countries, intestinal spirochaetosis rates are between 2% and 7%, between 11% and 34% in less developed countries, and up to 54% in homosexual men and HIV-infected patients (OR: 4.48)⁽⁵⁾, without being related to the viral load.

Most cases of intestinal colonization are asymptomatic and detected during a screening colonoscopy^(6,12). The symptoms are nonspecific: chronic watery diarrhea (most common clinical manifestation), abdominal pain, alternation between diarrhea and constipation, abdominal distension, rectal bleeding, and mucous anal discharges^(3,5,8,14). In cases of severe immunosuppression, cases of septic shock with associated spirochaetemia due to *B. pilosicoli*, with high mortality, have been described^(4,5,7).

During the colonoscopy study, the usual thing is identifying a normal-looking mucosa. However, findings such as mild erythema and pseudopolyps have been reported, which, in some cases, can cause severe acute inflammatory reactions with cryptic abscesses and ulcers, the latter seen in HIV-positive patients⁽⁶⁾. Given these unusual findings of such severity, other more prevalent infectious causes must be ruled out before making the diagnosis⁽⁶⁾.

The gold standard for diagnosis is histopathological study. Therefore, it is necessary to provide the pathologist with clear and complete clinical information on the clinical picture of chronic diarrhea with colonic mucosa with a normal endoscopic appearance so that the level of suspicion of this entity increases. Colonic biopsies show the presence of spirochaetes oriented perpendicularly on the luminal surface⁽¹²⁾. It can be frequently confused with mucin⁽¹⁰⁾; microscopy shows destruction of the microvilli and a mild inflammatory infiltrate in the lamina propria. Spirochaetes can be identified with hematoxylin and eosin staining, which allows seeing a basophilic band at the luminal pole of the epithelial cells that respects the surface of the goblet cells^(9,10,13,14). Another possible identification technique would be Warthin-Starry silver staining, which provides a better view of the spirochaetes and is the most specific technique^(7,10,14). In immunohistochemistry, *antitreponema pallidum* antibodies can be positive because there is a cross-reaction with the genus *Brachyspira*^(9,10). The causative species can be identified with polymerase chain reaction (PCR) amplification in feces or formalin-fixed tissue samples. When this last condition is not met, the sensitivity can be only 50%, which reduces its clinical usefulness and is reserved for research scenarios⁽¹⁰⁾.

Treatment is recommended if there are associated symptoms without other proven pathology; otherwise, an expectant attitude can be taken. Oral metronidazole is the medi-

cation considered the first choice and the most frequently used that results in the resolution of symptoms^(4,12,13). The doses are between 250 and 500 mg every 6 to 8 hours, lasting between 10 and 21 days. The most frequently used is 500 mg every 8 hours for 14 days. Other options are benzathine penicillin, doxycycline, and clindamycin^(5,8,12), although they frequently achieve partial control of symptoms. Resolution of symptoms may also occur without receiving any treatment.

It is unclear whether it is necessary to perform a follow-up colonoscopy and take new tissue samples for histopathological study to confirm the eradication of intestinal spirochaetosis.

Spirochaetosis is documented as an unusual cause of chronic diarrhea in the case reported. Its importance lies in

the fact that it must be considered in differential diagnoses, especially in patients with associated risk factors.

CONCLUSIONS

- Intestinal spirochaetosis is a cause of chronic diarrhea, mainly in patients with risk factors such as HIV infection, men who have sex with men, and ingestion of water contaminated with animal feces.
- There are no endoscopic findings characteristic of this disease.
- In most cases, the diagnosis depends mainly on the pathologist, who must suspect and identify it after knowing the clinical data of chronic diarrhea and normal colonoscopy.

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