Case Report

Colorectal spirochetosis

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ABSTRACT

Colorectal spirochetosis is a very rare pathologic condition that was been described several decades ago; however, its clinical significance is debatable in causing problems to human beings. We describe the first 2 documented cases of colorectal sprirochetosis in Saudi Arabia and discuss the different views about this entity and it's clinical significance.

Keywords: Colorectal, spirochetes, spirochetosis, infestation.

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It is well established that spirochetes may be found in the digestive tract of various species including humans;[1] however the pathologic potential and clinical significance is still controversial. We describe 2 cases of colorectal spirochetosis in a 16 year old male patient and a 40 year old male patient who were both known to have schistosomiasis and presented with perianal itching and abdominal pain with nausea and vomiting. Endoscopic examination was normal. Histological examination revealed large bowel mucosae that were covered by innumerable bacilli making a basophilic fringe over the brush border of the surface epithelium.

Case Report.

Patient 1. A 16 year old immunocompetent male patient known case of schistosomiasis on treatment presented with history of perianal itching for few weeks. His physical examination was normal. An initial work up was unremarkable. Procto-sigmoidoscopy was decided to document the presence of schistosoma ova. The endoscopic examination was not significant. Random biopsies were taken. Under the microscope, several fragments of rectal mucosa were seen, their surface epithelium was covered by several rod-like organisms making a basophilic fringe and sparing goblet cells (Figure 1). Warthin Starry stain highlighted the organisms (Figure 2). There is no significant associated mucosal inflammation. The patient was doing well after the endoscopy and received no treatment for the spirochetes. A few months later, he was symptom free.

Patient 2. A 40 year old immunocompotent male patient diagnosed to have schistosomiasis 25 years ago, presented with right upper quadrant pain aggravated by fatty meals and associated with nausea and vomiting. Upper endoscopy showed esophageal reflux grade 1. Upper abdominal ultrasound showed multiple small gall bladder stones. Colonoscopy was also carried out and was unremarkable. Random biopsies were taken from descending colon, sigmoid colon and rectum. Histological examination was more or less the same as Patient 1 with numerous thin rod-like organisms covering the surface. These organisms again proved to be positive with Warthin Starry stain. He received no treatment for the spirochetes; however, he underwent laparoscopic cholecystectomy and several months after the

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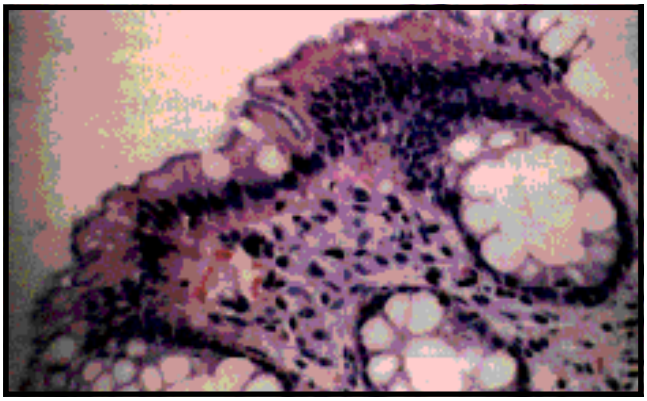

Figure 1 - High power view of colonic mucosa that is covered by a basophilic fringe formed by numerous spirochetes (Hematoxylin & Eosin original mangification x 400).

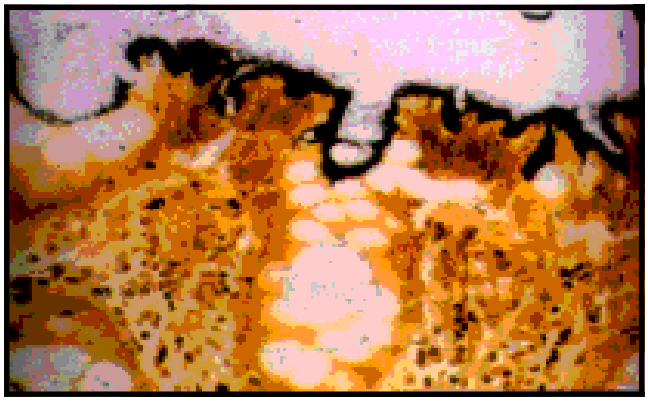

Figure 2 - The organisms (black) were highlighted by Warthin-Starry stain (Warthin-Starry, original magnification x 400).

surgery he was symptom free.

Discussion. Infestation of the human intestinal tract by spirochetes and spirochetes like organisms has been well known for more than a century. Several genera have been accused of this infestation including *Treponema, Borrelia, Leptospira, Serpulina, Anguillina* and *Brachyspira*. The 2 exceptional subspecies that have been claimed to colonize the intestinal tract and possibly cause diseases are *Brachyspira aalborgi* and *Anguillina coli*, the latter being more clinically significant than the former.[2] The incidence of spirochete infestation is variable and ranges from <1% up to 16.5%.[3,4] In an unpublished study by one of the authors, the incidence was found to be <2% in Saudi Arabia when approximately 110 cases of colorectal biopsies that were reported as normal were examined retrospectively, only 2 cases were positive for intestinal spirochetes. Although the presence of spirochetes in the intestinal tract has long been recognized, their clinicopathological implications are uncertain. In several cases, it has not been possible to relate the colonization of the enteric mucosa by spirochetes to gastrointestinal symptoms, accordingly, those microorganisms have been considered to be harmless commensals. In other cases spirochetes was the culprit in many gastrointestinal symptoms, such as diarrhea, constipation, rectal bleeding, abdominal pain and discharge.[5-7] And to complicate the issue further, therapeutic trials have contributed to the controversy over the pathogenicity of intestinal sprirochetes, some antibiotics have been accompanied by remission of symptoms in some cases but failed in others.[3] The route of infection is not clear. Since there is relatively a high prevalence of intestinal spirochetosis in homosexual men,[8] sexual contact has been put forward as a source of infection, adding to that, the immune deficiency status of some of these individuals has contributed to the symptoms caused by spirochetes, however, the discovery of intestinal spirochetes in children as well as immunocompetent persons defies the above claim that sexual contact is the source of infestation. Some other authors have suggested that the spirochetes may be enteric commensals that became opportunistic pathogens due to unknown factors, probably including alteration of local microenvironment.[9]

In our 2 cases, as well as in others described in the literature, there was associated infestation with other parasites, making it possible that some sort of a synergistic effect may have occurred between the two microorganisms.[10] It is possible that certain species of intestinal spirochetes are primary pathogens per se. Spirochete pathogenicity seems to depend on the extent and degree of infestation, regardless of the presence or absence of tissue damage. Some authors have hypothesized that the overgrowth of spirochetes and subsequent heavy colonization of the epithelium leads to symptoms. Others have suggested that clinical symptoms can also be secondary to an immune reaction elicited by the penetration of spirochetes in the mucosal cells and uptake by macrophages in the lamina propria.[1,5,9] In our cases, the presence of intestinal spirochetes was unrelated to the symptoms that the patient presented with, this is in keeping with the theory that most of the intestinal spirochetes are of no clinical significance. The other interesting aspect is that theses 2 cases represented the first case reports documented by histology in the area of Saudi Arabia, although discovered previously in another gulf state but still not seen until recently in Saudi Arabia.[11]

In conclusion, colo-rectal infestation by spirochetes in Saudi Arabia does occur, however, as in other regions, its clinical significance is debatable and most probably they represent harmless commensals.

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