

Research Article

Inter-Sigmoid Fistula in IBD- A Rare Phenomenon

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Abstract: **Introduction:** Fistulas occur in up to 50% of people with Crohn's disease. They are abnormal, tunnel-like connections that form between the intestine and other organs, or to the skin. The most common type is the perianal fistula (near the anus), which can cause pain, swelling, and discharge. It connects the anal canal to the skin near the anus and causes leakage of pus or stool and pain. The entero-enteric fistula connects one part of the bowel to another whereas entero-vesical is between bowel and bladder, which can lead to urinary tract infections (UTIs) and passing air/stool while urinating. The entero-cutaneous connects the bowel to the abdominal surface, draining fluid or stool to the skin and entero-vaginal connects the bowel to the vagina. **Case Report:** We report a thirty-six-year-old male who was diagnosed to be having ulcerative colitis twelve years back on basis of clinical presentation, colonoscopy and biopsy. He was started on oral mesalamine, immunosuppressants in form of oral azathioprine and steroids and other supportive therapy. He intermittently presented in flares and used to be given injectable steroids for five days. He was steroid dependent and, on an average, had 1-2 episodes of flare per year. After, that for few years he went in remission and stopped medications on his own. He remained asymptomatic for next 4 years but then suddenly went into flare, for which he was restarted on treatment but due to lack of full response, he was subjected to CECT Enterography which revealed multifocal areas of circumferential enhancing mural thickening in distal sigmoid colon and descending colon with luminal narrowing and surrounding fat stranding. A fistulous communication was seen between two adjacent segments of the distal sigmoid colon, suggestive of entero-enteric fistula. The rest CECT Enterography findings were within normal limits. The colonoscopy revealed suspected ulcero-proliferative growth at 15 cm from anal verge in sigmoid colon with multiple pseudo polyps in sigmoid colon. The histopathology from the suspected growth and pseudo polyps were negative for malignancy and showed dense inflammatory infiltrate, cryptitis and crypt abscess. There was no history of fever or blood in stools but patient was regularly passing mucus with stools on daily basis. There was no tenderness on abdominal examination or fistula at any other site. He is being planned for biological therapy and has no clinical and radiological findings suggestive of Koch's infection in body. **Conclusion-** IBD patients in 10 % cases in beginning can behave as indeterminate one but with time proceed either towards ulcerative colitis or crohn's disease. In our case, it started as ulcerative colitis and later developed mixed picture of crohn's as evidenced by fistula formation, in background of pseudo polyps, cryptitis and crypt abscess which goes in favour of ulcerative colitis. Moreover, inter-sigmoid colon fistula seen in our case, has rarely been reported in literature.

Keywords: Crohn's disease, Ulcerative colitis, Fistula, Sigmoid colon, Pseudo polyps, Skip lesions, Aphthae

INTRODUCTION

Inflammatory bowel disease (IBD) defines a category of chronic, relapsing, and remitting conditions characterized by sustained inflammation within the gastrointestinal tract. [1] The etiology of IBD is not fully understood. It is thought that several factors, such as infectious agents, race and ethnicity, immune system disorders, environmental factors, or a complex interplay of some/all of these factors, have been implicated as potential triggers for intestinal inflammation in IBD. [2-4] IBD is classified into two major subcategories, including ulcerative colitis (UC) and Crohn's disease (CD). Crohn's disease, represented by idiopathic transmural inflammation anywhere along the gastrointestinal (GI) tract, is on the rise in global incidence without known etiology. This inflammation manifests in inflammatory, stricturing, or increase the pressure with relative ischemia in the pre-stenotic dilated bowel loops which potentially leads to penetrating phenotypes including fistulas, abscesses and fissures. [5-

7] Crohn's disease is an inflammatory disease affecting the gastrointestinal (GI) tract and is characterized by persistent inflammation in the gut, which can lead to the formation of abnormal connections called fistulas. These fistulas can occur between the GI tract and the abdominal cavity, adjacent organs, or the skin. [8] The most common type is perianal fistula, which forms between the rectum and the skin near the anus. The exact mechanism of fistula formation is not fully understood but research suggests that factors such as epithelial to mesenchymal transition, matrix metalloproteinase, immune system dysregulation, and microbiota may contribute to their development. There is currently no definitive treatment for fistula closure, but options include surgery, endoscopic procedures, antibiotics, biologic agents and immunosuppressive drugs. These treatments can be used alone or in combination. However, recurrence is a significant challenge that needs to be addressed in the case of fistula treatment. [9]

MATERIALS AND METHODS

We report a thirty-six-year-old male who was diagnosed to be having ulcerative colitis twelve years back on basis of clinical presentation, colonoscopy and biopsy. He was started on oral mesalamine, immunosuppressants in form of oral azathioprine and steroids and other supportive therapy. He intermittently presented in flares and used to be given injectable steroids for five days. He was steroid dependent and, on an average, had 1-2 episodes of flare per year. After, that for few years he went in remission and stopped medications on his own. He remained asymptomatic for next 4 years but then suddenly went into flare, for which he was restarted on treatment but due to lack of full response, he was subjected to CECT Enterography which revealed multifocal areas of circumferential enhancing mural thickening in distal sigmoid colon and descending colon with luminal

narrowing and surrounding fat stranding. A fistulous communication was seen between two adjacent segments of the distal sigmoid colon, suggestive of entero-enteric fistula. The rest CECT Enterography findings were within normal limits. The colonoscopy revealed suspected ulcero-proliferative growth at 15 cm from anal verge in sigmoid colon with multiple pseudo polyps in sigmoid colon. The histopathology from the suspected growth and pseudo polyps were negative for malignancy and showed dense inflammatory infiltrate, cryptitis and crypt abscess. There was no history of fever or blood in stools but patient was regularly passing mucus with stools on daily basis. There was no tenderness on abdominal examination or fistula at any other site. He is being planned for biological therapy and has no clinical and radiological findings suggestive of Koch's infection in body.

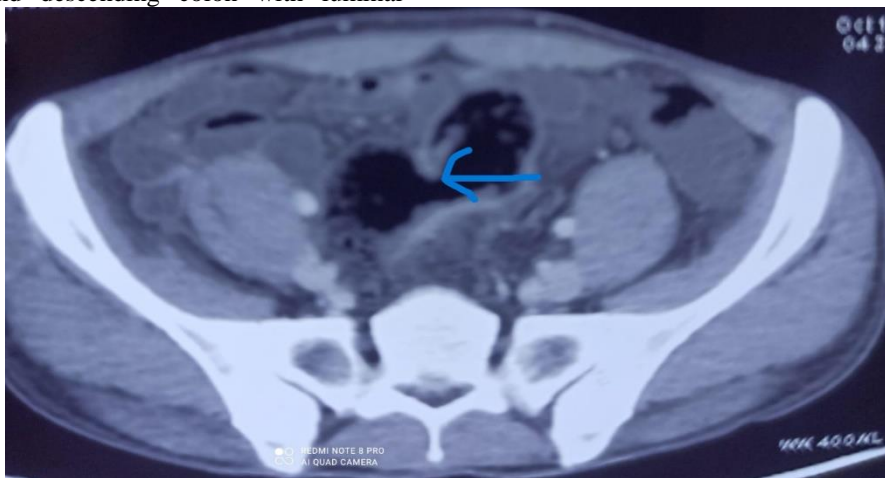


Figure 1- CT Enterography Showing Inter-Sigmoid Colon Fistula (Blue arrow)

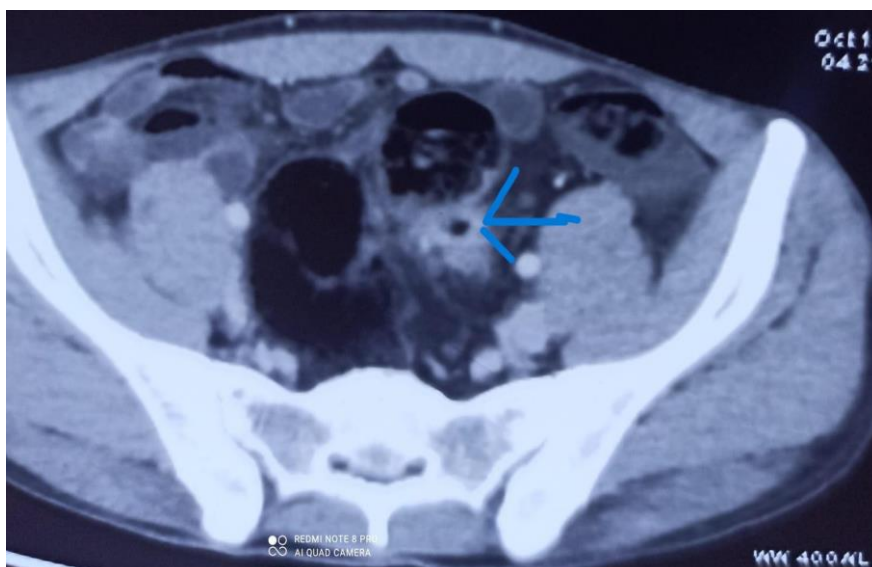


Figure 2- CT Enterography showing trapped air in sigmoid colon loop (Blue arrow)



Figure 3- Colonoscopy showing multiple Pseudo polyps in sigmoid colon favoring Ulcerative colitis



Figure 4- Colonoscopy showing multiple aphthae favoring crohn's disease



Figure 5- Colonoscopy showing normal rectal mucosa favoring crohn's disease

DISCUSSION

It is well known fact that 10 % of cases behave as indeterminate colitis in beginning of disease and with passage of time, settle either towards crohn's or ulcerative colitis. Our case is exception in the sense, that it started as clear cut case of ulcerative colitis in beginning which was diagnosed on basis of colonoscopy findings and histopathological evidence of cryptitis and crypt abscess. Later, on it developed mixed features of both UC and crohn's. Pseudo polyps and repeat histopathological evidence of cryptitis and crypt abscess favors UC but rectal sparing, skip lesions, multiple aphthae and inter-sigmoid fistula favors crohn's disease. Moreover, inter-sigmoid fistula is very rare because most common fistula in crohn's are peri-anal and entero-

enteric fistula are usually between ileum and sigmoid colon.

CONCLUSION

IBD patients in 10 % cases in beginning can behave as indeterminate one but with time proceed either towards ulcerative colitis or crohn's disease. In our case, it started as ulcerative colitis and later developed mixed picture of crohn's as evidenced by fistula formation, in background of pseudo polyps, cryptitis and crypt abscess which goes in favor of ulcerative colitis. Moreover, inter-sigmoid colon fistula seen in our case, has rarely been reported in literature.

Conflict of Interest- No conflict of interest or financial support was taken and prior permission from patient and relatives was taken before publishing the case report.

to its severe adverse events: true or false? *Cell J.* 2020;22(Suppl 1):1–18. doi: 10.22074/cellj.2020.7603

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