J MEDICINE 2024; 25: 83-86

Case Report

A Young Adult with Crohn's Arthritis Necessitating Upper Gastrointestinal Tract Surgery - A Case Report

Tasmina Chowdhury[1], Israt Jahan Urmee[2], Khan Abul Kalam Azad[3]

Abstract

Upper gastrointestinal tract predominant Crohn's Disease (CD) remains an uncommon clinical entity, manifesting limited or vague symptomatology, thus delaying clinical suspicion and subsequent diagnostic evaluation. For that reason, it has not been widely described and there is a lack of clear recommendations for diagnosis or management. Standard Inflammatory Bowel Disease (IBD) evaluation including serologic testing, imaging, and endoscopy may initially not be fruitful. Furthermore, endoscopic evaluation may be inconclusive even in patients with long standing-disease. We describe a 30-year-old male who suffered long with persistent iron-deficiency anemia (IDA) and occasional melaena along with predominant axial arthropathy. Extensive in-patient testing including multiple endoscopic evaluation with standard biopsy were unfruitful. Ultimately, long standing recurrent gastric and duodenal ulcer, suggestive colonoscopy finding, and positive fecal calprotectin in a patient along with spondyloarthropathy drove us toward the diagnosis of Crohn's disease. We started steroid, PPI, sulfasalazine and the patient is on follow up.

Key words: *Gastrodudenal crohn's disease, Crohn's arthritis, iron-deficiency anemia (IDA)*

DOI: *https://doi.org/10.3329/jom.v25i1.70532*

Received: *8.12.2023* ***Accepted:*** *18.12.2023*

Introduction:

Crohn's disease (CD) is a heterogeneous disorder with multifactorial etiology, including genetic factors, environmental insults and intestinal microbiota, characterized by chronic, segmental and transmural inflammation that affects the gastrointestinal tract and may involve any segment of the oral cavity up to the anus.[1] It is most often associated with involvement of the terminal ileum or colon and may rarely affect the stomach and duodenum.[2] CD confined to the stomach or duodenum without involvement of the small intestine and colon is very rare.[2] The first report of duodenal involvement of CD was described by Gottlieb and Alpert in 1937, and Ross reported gastric CD for the first time in 1949.[3] Enteropathic arthritis (EA) is a spondyloarthritis (SpA) which occurs in patients with inflammatory bowel diseases (IBDs) and other gastrointestinal diseases. There is no gold standard criteria for the diagnosis of CD, The diagnosis depends on clinical, endoscopic, histological and radiological evaluation. Here we discuss on a 30-year-old male who required gastrojejunostomy , got treatment of PUD and had a long suffering without any definitive diagnosis and improvement of his IDA and spondyloarthropathy. Clinical suspicion on atypical CD, correlation with arthritis, further evaluation through history and investigation led the diagnosis of Crohn's arthritis.

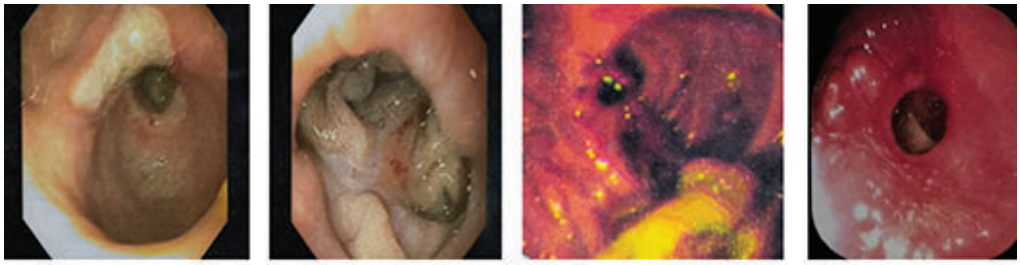
Case Summary:

A 30-year-old man street food vender, gave history of undergoing gastrojejunostomy due to gastric outlet obstruction 14 years back, while he presented with severe vomiting and constipation. He remained symptom free for about 1 year, since then he has been suffering from recurrent episodes of weakness and fatigue. He also had few episodes of melaena, occasional short lived mild postprandial abdominal pain and painful oral ulcer. He took short course

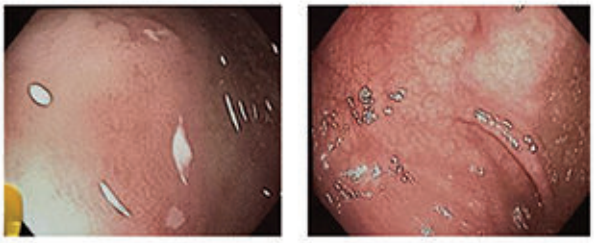
1. Assistant Professor, Dept. of Medicine, Popular Medical College & Hospital(PMCH)
2. Assistant registrar, dept of Medicine, Popular Medical College & Hospital (PMCH)
3. Professor of Medicine, Principal, PMCH

Corresponding author: Dr. Tasmina Chowdhury, Assistant Professor, Dept of Medicine, Popular Medical College & Hospital (PMCH), email: tasminac7@gmail.com, Phone: 01716415300

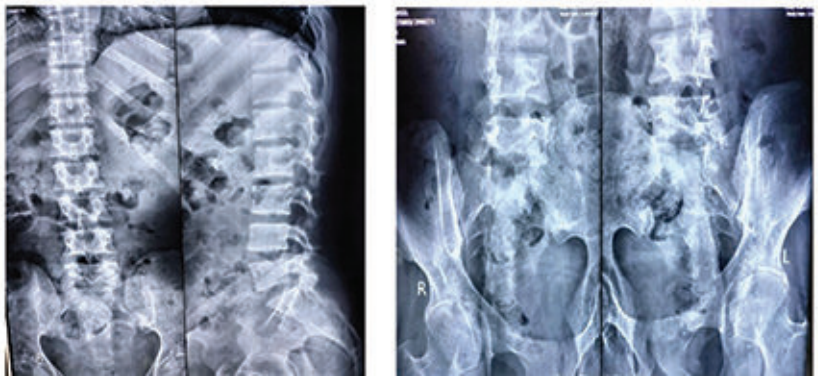
NSAIDs on few occasions for last 10 years. He denied any history suggestive of liver disease. Other than episodes of melaena his bowel habit was almost normal. Every time he visited doctor found to have severe iron deficiency anaemia. He got repeated admission including tertiary care hospital, went through series of investigations including Hb electrophoresis, bone marrow study and recurrent endoscopy. He was found to have bleeding PUD and received more than 20 units of blood transfusion, H Pylori eradication therapy, PPI in multiple settings in last 13 years. For about last ten years he developed recurrent episodes of inflammatory joint pain involving his both knees, wrists, back and occasionally small joints of both hands. Gradually his pain became persistent increased in intensity, became confined to neck, back and waist. He denied dysuria, red eye, skin rash, breathlessness or dry cough. He had gross weight loss. On general examination he was moderately anaemic had generalized muscle wasting and lumbar lordosis. His cervical, thoracic and lumbar spinal movement was restricted and painful. Modified Shober's test was positive. Features of sacroiliitis were present. There was no feature of peripheral joint involvement or enthesitis. We considered the differentials of spondyloarthropathy with NSAIDs induced bleeding PUD and Crohn's disease with enteropathic arthritis. His previous investigations revealed microcytic hypochromic anemia (Hb level ranging from 7-7.4 g/dl and MCV: 69.2-71 fl and MCHC: 19-20 pg) with a normal total and differential leucocyte count; PBF showed moderate microcytic hypochromic anemia. Iron profile showed low Iron and Ferritin level and raised TIBC. Bone marrow study revealed micro normoblastic erythroid hyperplasia. Hb electrophoresis was normal. RA, Anti-CCP Ab were negative. Faecal calprotectin was borderline high 86mcg/g (<80 mcg/g normal).Stool occult blood test was positive and Stool R/M/E showed Pus cell - 0-2/HPF, RBC- 4-6/HPF. CRP: 8.6 mg/l. Previous Endoscopy of upper GIT on different occasions revealed active ulcer with bleeding. This time Colonoscopy with biopsy showed multiple aphthoid ulcers in the terminal ileum. Histopathological findings were consistent with active colitis as lamina propria revealed infiltrate of acute and chronic inflammatory cells and no granuloma was found. X-ray of lumbosacral spine and SI joints showed lumbar lordosis with sclerosis of SI joints.



Stomal ulcer Bleeding PUD



Aphthoid ulcers on terminal ileum



Lumbar lordosis Sclerosis of SI joints

Figure:

Discussion:

CD is almost always found in the small intestine and colon.[4] CD may rarely affect the stomach and duodenum, and the frequency of gastroduodenal CD is reported to range between 0.5% and 4.0% in all patients with CD.[4,5] Most patients with gastroduodenal CD have concomitant lesions in the terminal ileum or colon[6]. Many patients with gastroduodenal CD are asymptomatic. [4, 5,7] The most common symptom is epigastric pain.[4,5]Although hematemesis and melena may rarely occur, upper gastrointestinal bleeding may manifest in the form of chronic anemia.[2] A cross-sectional study has demonstrated that proximal CD affects younger, nonsmoker patients, is less likely to involve the colon, is more frequently present with concomitant ileal involvement and stenosing behavior, and has a higher probability of having one or more abdominal surgeries. The most common complication of gastroduodenal CD is gastric outlet obstruction due to stricture formation.[8,9] In contrast to peptic ulcer disease, perforation and severe gastrointestinal hemorrhage are seldom detected.[5] The diagnosis of gastroduodenal CD requires a high level of clinical suspicion.[5] Identification of granulomas on biopsy is the most important step in confirming upper CD, but granulomas are found on endoscopic biopsies only between 0% and 83% of the time, and focally distributed nonspecific inflammation is a more common finding on histology.[8] Thus, if there are no definite histologic findings, gastroduodenal CD can still be diagnosed by comprehensive clinical evaluation including detailed history taking, physical examination, radiologic findings, endoscopic evaluation, and histologic and laboratory investigations.[10] Inflammatory arthritis is the most common EIM of IBD, with a prevalence 6%–46%.[11] Interestingly, articular alterations can be diagnosed before, simultaneously, or after the diagnosis of IBD. It can be peripheral, axial, or both, as commonly seen in other types of SpA.[12] The axial involvement is found to be present in 2%–16% of IBD patients, with a higher prevalence in CD patients than in UC ones. Moreover, the prevalence of sacroiliitis (asymptomatic and symptomatic) is between 12% and 20%.[13] Our patient presented with the feature of acute abdomen due to GOO at his age of sixteen. Since then suffering from upper GI bleeding, IDA and thereafter from spondyloarthropathy. Unlike The common features of CD he had insignificant abdominal pain, no altered bowel habit, diarrhoea or per rectal bleeding. Endoscopy in multiple setting showed ulcers in stomach, duodenum, and stoma. He was treated as a case of bleeding peptic ulcer disease and spondyloarthrhopathy was considered as separate entity. Long standing active PUD in a young adult despite of taking repeated H pylori therapy and PPI is unlikely. He used to take occasional NSAID, but his bleeding episodes started before he had started taking NSAIDs. CD can produce such ulcers and bleeding which aggravate due to NSAID. The total scenario provoked high clinical suspicion of CD and we proceeded for further search. His recent colonoscopy showed multiple aphthoid ulcer in terminal ileum which is a likely feature of CD. Though biopsy showed no granuloma, it is not unusual as we discussed above. Moreover our patient had spondyloarthropathy typical of enteropathic arthritis. Considering all, it was an atypical case of upper gastrodudenal CD along with silent ileal involvement and enteropathic arthritis. The first line of the treatment in active gastroduodenal CD is a proton pump inhibitor in combination with steroids.[14] However, surgical intervention is required when there are complications such as GOO, bleeding, perforation and fistula formation.[15] Glucocorticoids can be used in management of both IBD and arthritis.[16] According to 2019 ACR guidelines for management of active ankylosing spondylitis, sulfasalazine should be considered only in patients with prominent peripheral arthritis.[17] TNF-á inhibitors are proven to have therapeutic benefit for SpA management according to the pivotal phase III trials. These TNF-á inhibitors are infliximab, etanercept, adalimumab, certolizumab, and golimumab.[16] Treatment was started with PPI, oral steroid and sulfasalazine and patient is on follow up.

Conclusion

The prevalence, complications, therapeutic and prognostic implications of involvement of the esophagus, stomach and duodenum in CD has not been thoroughly elucidated to date. Endoscopy and histological study are useful diagnostic methods for the adequate identification and characterization of the lesions that can affect these segments, but these results should be interpreted with caution, staying aware of other conditions that can mimic this disease. This rare case highlights the importance of clinical suspicion and comprehensive clinical evaluation for the diagnosis of isolated gastrodudenal CD.

References

1. Gajendran M, Loganathan P, Catinella AP, Hashash JG. A comprehensive review and update on Crohn's disease. Dis Mon. 2018;64:20–57. [PubMed] [Google Scholar]

2. Kefalas CH. Gastroduodenal Crohn's disease. Proc (Bayl Univ Med Cent) 2003;16:147–151. [PMC free article] [PubMed] [Google Scholar]

3. Hwang YY, Jo YK, Cha JM, Kim KH, Na IH, Lee DH, Ha HS, Kim SH, and Song CS. A Case of Crohn's Disease Confined to the Stomach. Korean J Gastrointest Endosc. 2003;26:146–149. [Google Scholar]

4. Ingle SB, Adgaonkar BD, Jamadar NP, Siddiqui S, Hinge CR. Crohn's disease with gastroduodenal involvement: Diagnostic approach. World J Clin Cases. 2015;3:479–483. [PMC free article] [PubMed] [Google Scholar]

5. Kefalas CH. Gastroduodenal Crohn's disease. Proc (Bayl Univ Med Cent) 2003;16:147–151. [PMC free article] [PubMed] [Google Scholar]

6. Henter JI, Horne AC, Aricó M, et al. HLH 2004: Diagnostic and therapeutic guidelines for hemophagocytic lymphohistiocytosis. Pediatr Blood Cancer. 2007;48(2):124 131. [PubMed] [Google Scholar]

7. Loftus EV Jr. Upper gastrointestinal tract Crohn's disease. Clin Perspect Gastroenterol. 2002;5:188–191. [Google Scholar]

8. Isaacs KL. Upper gastrointestinal tract endoscopy in inflammatory bowel disease. Gastrointest Endosc Clin N Am. 2002;12:451–462, vii. [PubMed] [Google Scholar]

9. Yamamoto T, Allan RN, Keighley MR. An audit of gastroduodenal Crohn disease: clinicopathologic features and management. Scand J Gastroenterol. 1999;34:1019–1024. [PubMed] [Google Scholar

10. Ye BD, Jang BI, Jeen YT, Lee KM, Kim JS, Yang SK. [Diagnostic guideline of Crohn's disease] Korean J Gastroenterol. 2009;53:161–176. [PubMed] [Google Scholar]

11. Colia R, Corrado A, Cantatore FP. Rheumatologic and Extraintestinal Manifestations of Inflammatory Bowel Diseases. Ann Med. 2016;48:577–585.10.1080/07853890.2016.1195011Search in Google Scholar PubMed

12. Firestein GS, Budd RC, Gabriel SE, et al. Firestein & Kelley's Textbook of Rheumatology, 11th edn. Philadelphia. Elsevier, 2021.Search in Google Scholar

13. Gionchetti P, Calabrese C, Rizzello F. Inflammatory Bowel Diseases and Spondyloarthropathies. J Rheumatol Suppl. 2015;93:21–23.10.3899/jrheum.150628Search in Google Scholar

14. Goyal J, Khan AS, Goel A, Weber F. Isolated gastroduodenal Crohn's disease in a septuagenarian man. Endoscopy. 2014;46 Suppl 1 UCTN:E167–E168. [PubMed] [Google Scholar]

15. Yamamoto T, Allan RN, Keighley MR. An audit of gastroduodenal Crohn disease: clinicopathologic features and management. Scand J Gastroenterol. 1999;34:1019–1024. [PubMed]

16. Firestein GS, Budd RC, Gabriel SE, et al. Firestein & Kelley's Textbook of Rheumatology, 11th edn. Philadelphia. Elsevier, 2021.Search in Google Scholar

17. Ward MM, Deodhar A, Gensler LS, et al. 2019 Update of the American College of Rheumatology/Spondylitis Association of America/Spondyloarthritis Research and Treatment Network Recommendations for the Treatment of Ankylosing Spondylitis and Nonradiographic Axial Spondyloarthritis (in Eng). Arthritis Rheumatol. 2019;71;1599–1613.10.1002/art.41042Search in Google Scholar