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. It is most often associated with involvement of the terminal ileum or colon and may rarely affect the stomach and duodenum. CD confined to the stomach or duodenum without involvement of the small intestine and colon is very rare. 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. Enteropathic arthritis (EA) is a spondyloarthitis (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 suffered long with persistent iron-deficiency anemia (IDA) and occasional melena 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.

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 melena, occasional short lived mild postprandial abdominal pain and painful oral ulcer. He took short course
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.

Figure:
Discussion:
CD is almost always found in the small intestine and colon. 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. Most patients with gastroduodenal CD have concomitant lesions in the terminal ileum or colon. Many patients with gastroduodenal CD are asymptomatic. The most common symptom is epigastric pain. Although hematemesis and melena may rarely occur, upper gastrointestinal bleeding may manifest in the form of chronic anemia. 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. In contrast to peptic ulcer disease, perforation and severe gastrointestinal hemorrhage are seldom detected. The diagnosis of gastroduodenal CD requires a high level of clinical suspicion. 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. 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. Inflammatory arthritis is the most common EIM of IBD, with a prevalence 6%–46%. 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. 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%. 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, diarrhea 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 spondyloarthropathy 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 gastroduodenal 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. However, surgical intervention is required when there are complications such as GOO, bleeding, perforation and fistula formation. Glucocorticoids can be used in management of both IBD and arthritis. According to 2019 ACR guidelines for management of active ankylosing spondylitis, sulfasalazine should be considered only in patients with prominent peripheral arthritis. 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. 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 gastroduodenal CD.

References


