Crohn's disease in a patient with pre-existing Ankylosing Spondylitis- A case report

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Abstract

Crohn's disease is an inflammatory bowel disease which may be present in association with Ankylosing Spondylitis. Here, we are presenting a case of 31-year-old male who presented to us with abdominal pain and unexplained anaemia at Sheikh Russel National Gastroliver Institute and Hospital, Dhaka in 2022. . He was previously diagnosed as Ankylosing Spondylitis. With the help of history, physical examination, endoscopic immunological and radiological examination he has been diagnosed as a case of Crohn's disease along with pre-existing Ankylosing Spondylitis.

Keywords: Ankylosing Spondylitis, Crohn's disease.

Introduction:

The diagnostic criteria of Ankylosing Spondylitis (AS) are in accordance to the modified New York criteria that include both clinical and radiological aspects.[1]The spondyloarthritis includes sacroiliitis, inflammatory back pain, asymmetric oligoarthritis predominantly of the lower limbs, enthesitis, dactylitis and extra-articular manifestations such as gut inflammation, psoriasis and anterior uveitis. Clinical criteria included: low back pain and stiffness for >3 months, which improve with exercise and not relieved by rest, limitation of motion of the lumbar spine in both the sagittal and frontal planes, and restriction of chest expansion relative to normal values corrected for age and sex. Radiological criterion is bilateral sacroiliitis grade ≥2 or unilateral sacroiliitis grade 3 to 4. A definite AS is yielded by the radiological criterion and at least 1 clinical criterion.[2,3]

The diagnostic criteria of Crohn's Disease (CD) include the following aspects:a history of abdominal pain, vomiting, diarrhea, weight loss or rectal bleeding; radiologic findings of stricture or fistula formation, mucosal cobble stoning, or deep ulceration; endoscopic appearances of cobble stoning, linear ulceration, or skip lesions; and pathologic confirmations of transmural inflammation or noncaseating epithelioid granulomas.[4]

The Patient having CD with Extra Intestinal Manifestations (EIM) has been associated with increased morbidity and worse quality of life compared to their counterparts without EIMs.[5,6] Several studies have estimated the occurrence of Spondyloarthropathies (SpA) in patients with IBD ranging between 1-15% confirming that SpA is the most frequent extra-intestinal manifestation in patients with IBD.[6-11]

Case Presentation:

A 31 year gentleman having Ankylosing spondylitis for 15 years presented with complaints of recurrent episodes of abdominal pain and repeated blood transfusion for the last 1 year. The pain was located at periumblical region, sudden in onset, mild to moderate intensity, cramping in nature without radiation, aggravated after meal and relieved by oral medication and/or induced vomiting. Each episode of pain occurred 2-3 days apart and persisted for 3-7 hours. Pain was associated with vomiting which occurs 2/3 hours after meal. Vomitus contains partially digested bile stained

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food particles but not mixed with blood. During attack of pain, he did not notice abdominal distension or any visible lump. He had history of alteration of bowel habit for same duration in the form of loose stool. Loose stool occurred 3/4 times a day and continued for 2/3 days with 2/3 weeks interval. Amount of stool was scanty, liquid in consistency (Bristol stool chart: Type 6). The patient occasionally passed black tarry stool for the last 3 months. He gave no history of steatorrhoea. Loose motion was not related to milk or other foods. He also complained of weight loss about 12 kg over last 8 months. Weight loss was not associated with anorexia but he was afraid of taking food as abdominal pain was aggravatedafter meal.This patient had no history of fever, cough, skin rash, red eyes, oral ulcer, night sweats,heat intolerance, tremor, any contact with known TB patient. His bladder habit was normal.

He was diagnosed as Ankylosing spondylitis 15 years back at the age of 16 years on the basis of inflammatory type of back pain, asymmetrical oligoarthritis of large joints with relevant investigations. He suffered from GBS (Guillain Barre Syndrome) 4 years back; now on recovery state with some degree of weakness in both lower limbs.

He visited physician for several times and hospitalized only for blood transfusion not for abdominal pain/or loose stool. He got physiotherapy for residual weakness of GBS and 7 units of blood transfusion from December 2020 to January 2021.

He is 5th issue of non-consanguineous parents. All of his brothers are in good health. There is no such type of illness in his family. No family history of GI malignancy, IBD and liver diseases.His maternal uncle had similar type of spinal gesture problem.

He is non-smoker, non-alcoholic and nor having history of taking betel leaf and nut. He belonged to a middle class family.He completed graduation in political science. He has medicine business. He was prescribed Sulfasalzine and NSAIDs for ankylosing spondylitis. He took Sulfasalzine irregularly and stopped it after taking for initial 4 years. But he has been using diclofenac in the form of tablets occasionally to relieve back pain. He often takes Ciprofloxacin and Metronidazole in the tablet form for relieving loose motion. Recently he has taken anti-H pylori medication but his symptoms persisted.

On physical examination, the patient looked ill having BMI-17.He was anaemic. The power of the lower limbs decreased and Schober's test positive with restrictive body movement.Rest of the general and systemic examinations were found normal.

His initial investigations after admission were as follows: Haemoglobin 6.1 gm/dL with low MCV and MCH, RDW CV-23%, Neutrophil-68%, Lymphocyte-22%, Monocytes-07%, Eosinophil -03%, Platelet count-8,85,000/mL, ESR-58, RBS-6.4, S.creatinine -1.03 mg/dl, CRP - 48mg/dL, SGPT- 21 U/L , S. Albumin-3.15 gm/dL, S. TSH- 2.75 pg/mL, S. Electrolytes, S.lipase are within normal range, HLA B27 – Positive, Occult blood test-Positive, fecal calprotectin-1105 mcg/mg, MT- Negative, .

X-rays

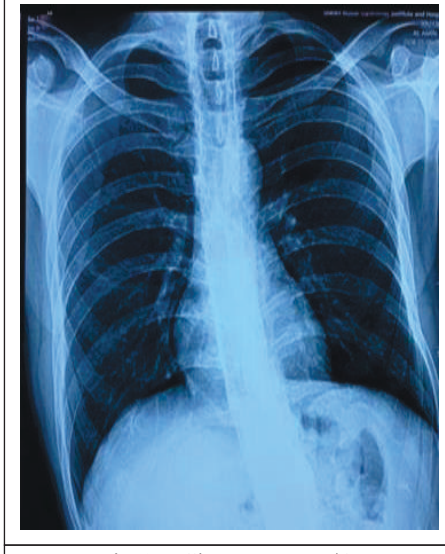


Fig 1a: Chest X-ray P/A:

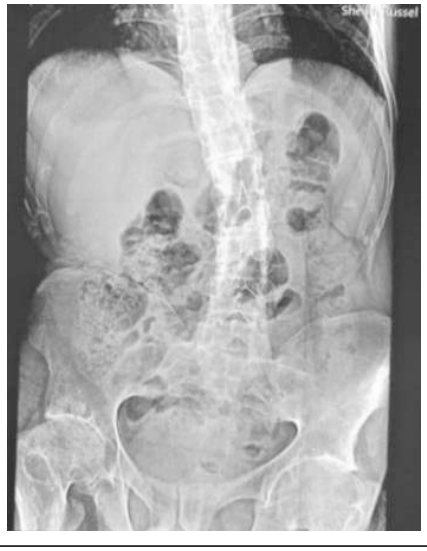


Fig 1b: X-ray of spine

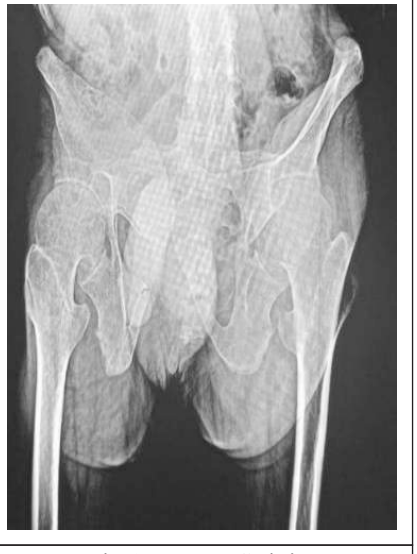


Fig 1c: X-ray SI joint

Chest X-ray was Normal **(**Fig 1a**)**, X-ray spine showed Bamboo spine and Scoliosis **(**Fig 1b**)** and X-ray of SI joints showed Sacroiliitis **(**Fig 1c**)**.

Endoscopy of upper GI tract

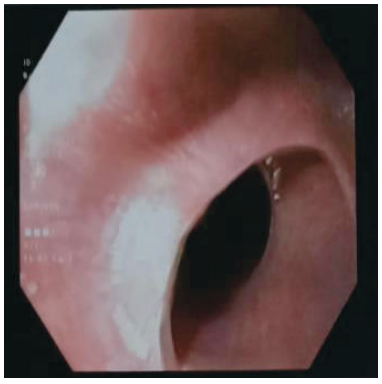


Fig 2a: Oesophageal web

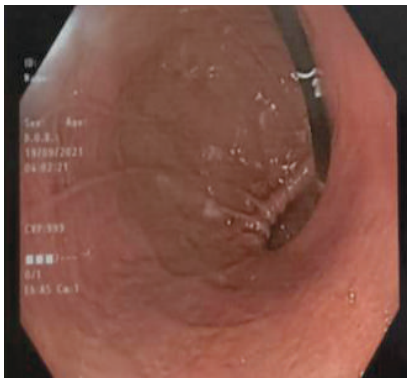


Fig 2b: Hiatus hernia

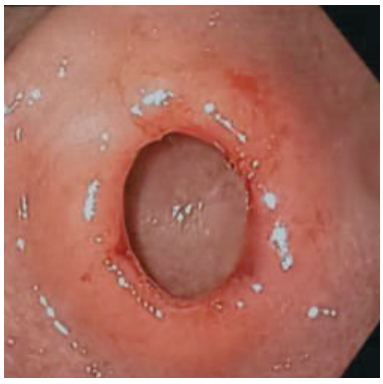
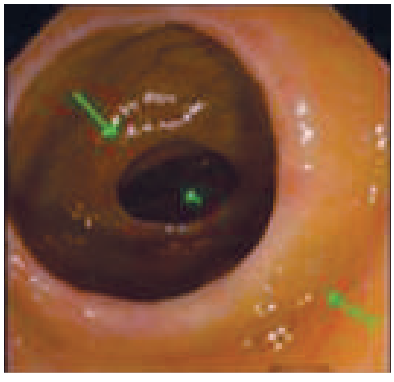


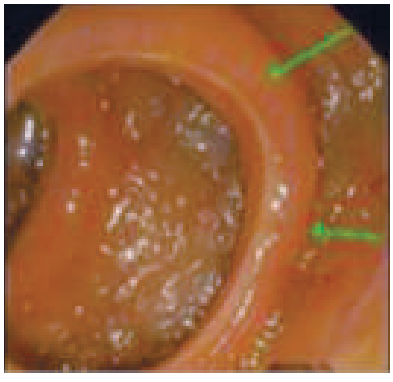
Fig 2c: Ulcer at rim of pylorus

It showed oesophageal web **(**Fig 2a), hiatus hernia **(**Fig 2b), and gastric ulcer with deformed bulb **(**Fig 2c**).** Biopsy was taken from the ulcers at stomach which revealed gastritis**.**

Single Balloon Enteroscopy

Multiple ring like ulcers (Fig 3) at mid ileum.





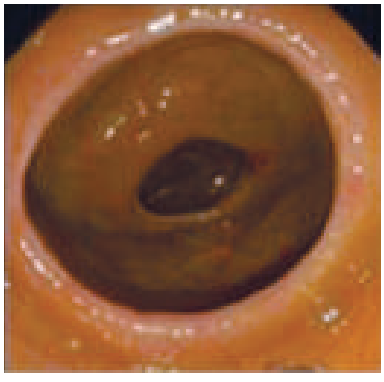
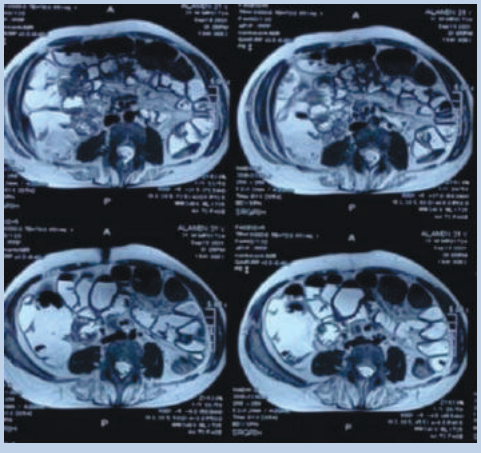


Fig 3: Single balloon enteroscopy showing multiple ring like circular ulcer and partial narrowing of the lumen



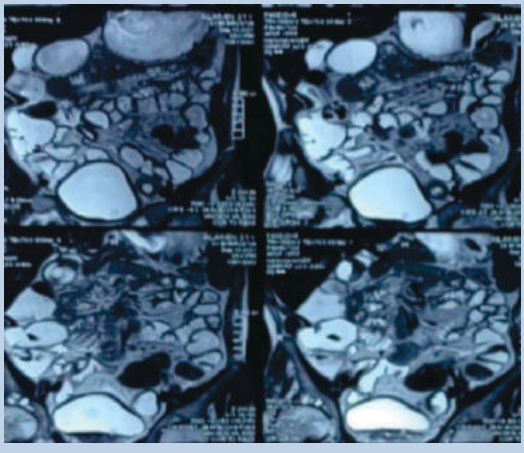


Fig 4:MR enterography shows mid ileal luminal narrowing and stricute

On the basis of history, clinical examination and relevant investigation, the patient is diagnosed as Crohn's Disease with Ankylosing spondylitis.

Discussion:

Concurrent incidence of Crohn's disease and Ankylosing spondylitis are not uncommon. It varies in different part of the world ranges from 1-15%.[6,7,8,9,10,11]In our case, the patient presented oligoarthritis and diagnosed as Ankylosing spondylitis earlier. After long 15 years later he had developed loose motion, abdominal pain, anaemia and diagnosed as Crohn's Disease. Song Liu et al described the symptoms of AS appeared earlier than CD in majority of patients, suggesting that CD is presumptively secondary to AS.[12]

The type of CD of this patient according to the Montreal classification is A2,L1+ L4,B2. A stands for the age of the patient. Age below 17 is A1, Age 17-40 is A2 and age above 40 will be titled as A3. The disease location is classified into L1 (terminal ileum), L2 (colon), L3 (ileo-colon), and L4 (upper gastrointestinal tract). The disease behavior is classified into B1 (inflammatory),B2 (stricturing), B3 (penetrating), and P (perianal lesion).Worth mentioning, the presence of chronic ileal lesions might be apredictor of an aggressive evolution of the spondyloarthropathy (SpA).[13]

Our patient presented with recurrent episodes of abdominal pain for 1 year which was similar to the data shown by Perler et al. According to that study, the most common presenting symptoms in CD found based on disease location at time of diagnoses were abdominal pain (82.14%) and tiredness/fatigue (72.41%) for ileal CD.[14]

Typical endoscopic findings in CD included discontinuous distribution of longitudinal ulcers (defined as ≥4 to 5 cm ulcers in the Japanese criteria), cobblestone appearance, and/or small aphthous ulcerations arranged in a longitudinal fashion.[15]In our patient, multiple ring like ulcers at mid ileum was found with histopathology showing active and chronic inflammation.

Fecal calprotectin (FC) is a biological marker of intestinal inflammation which was present in this patient. FC is one of the major proteins found in the cytosol of inflammatory cells.[16]The utility of FC as a surrogatemarker for endoscopic lesions in adults with IBD and found that cut-off value > 250 µg/g yielded sensitivity 71% and specificity 100% in distinguishing active and not active endoscopic mucosal disease.[17]In our case we found calprotectin level of 1105 mcg/gm which indicates active CD.

MR enterography is more effective than ultrasonogram in the evaluation of gastrointestinal tract, perianal region and complications in case of Crohn's disease.[18]Moreover, Lee et al have demonstrated that the effectiveness of MR enterography is comparable to that of CT enterography and also it has the advantage of not using ionizing radiations.[19]Aphthoid and deep ulceration, wall thickening (greater than 4 mm), intramural and mesenteric edema, stratified enhancement pattern of bowel wall, increased mesenteric vascularity, reactive lymphadenopathy are the common findings in MR Enterography in active Crohn's disease.[20] In MR Enterography, we found two short segment mild bowel wall thickening, mild luminal narrow ingand stricture formation at mid ileum and minimally englarged lymph nodes without central necorsis in our case.

IBD, however, precedes the development of spondylitis in most cases. On the other hand, a small proportion (<5%) of patients with established spondyloarthropathy can develop classic IBD within 10 years of the primary diagnosis: 80% of patients develop Crohn's disease and 20% ulcerativecolitis.[21] Same thing was found in our case. Patient was previously diagnosed as Ankylosing Spondylitis and now diagnosed as Crohn's disease.

It could be speculated that SpA and CD probably should be considered as distinct phenotypes of a common immune mediated inflammatory disease pathway rather than as separate disease entities and that ileitis of SpA might actually represent subclinical Crohn's disease.[22]

Treatment of overlapping Crohn's disease and Ankylosing spondylitis is not satisfactory sometimes. Low doses of systemic steroids and selective COXIBs may be used as a "bridge therapy" to oral SSZ (2–3 g/day). Inadequate response or intolerance to SSZ, biologic treatment (specific anti-TNFα) should be started.[23,24]

Conclusion:

Simultaneous presence of Ankylosing Spondylitis and Crohn's disease is rare. But they can overlap as both have immune mediated pathophysiology. So, extensive investigation is needed to diagnose vice versa.

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