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Case report

Eosinophilic enteritis with enteroliths: A diagnostic dilemma

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ABSTRACT

Introduction and importance: Eosinophilic gastroenteritis (EG) is a rare disease, characterized by eosinophilic infiltration of different layers of intestinal wall. Thus having a wide spectrum of presentation leading to diagnostic dilemma.

Case presentation: We report a case of 55 years old female who presented with pain in abdomen, constipation, vomiting off and on with passage of stools on alternate days since 6 months. Plain radiographs showed radio-opaque densities in abdomen. Contrast enhanced computed tomography (CECT) of abdomen showed Ileal stricture with dilated proximal bowel loops with enteroliths. Exploratory laparotomy confirmed ileal stricture with thickening of the mesentery and an ileal diverticulum. Resection of ileal stricture was performed. The resected segment contained seven hard, black enteroliths. Histopathology of the resected specimen confirmed EG. Stone analysis showed dense faecal matter with bile salts.

Clinical discussion: EG leads to symptoms ranging from vomiting, abdominal pain, diarrhoea, blood loss in stools, anaemia to malabsorption resulting in diagnostic dilemma. It may cause gastrointestinal obstructive symptoms secondary to stricture, depending upon the predominant layer involved.

Conclusion: The differential diagnosis of EG should always be considered when dealing with gastroenteritis presenting with radio-opaque densities in abdomen.

1. Introduction

The work has been reported in line with SCARE criteria [1]. Eosinophilic gastroenteritis (EG) is a rare inflammatory disease, presenting with wide spectrum of symptoms similar with other gastro-intestinal (GI) disorders. EG has been under diagnosed and considered a rare disorder. Eosinophilic gastroenteritis is defined as a disorder that selectively affects the gastrointestinal tract with eosinophil rich inflammation in the absence of any known causes for eosinophilia [2]. EG was first described by Kaijser in 1937 in two patients with syphilis who were allergic to neoarsphenamine [3]. The incidence of EG is estimated to be 1–30/100000, though accurate epidemiologic data is not available [4]. Patient with EG presents clinical features ranging from nausea, vomiting, abdominal pain, diarrhoea, haemorrhage and obstruction depending upon the predominant layer involving eosinophilic infiltration, and hence classified as mucosal, muscularis, and subserosal [5]. Involvement of mucosa and muscularis layer may result in stricture causing bowel obstruction [6]. Enterolithiasis was first described by Pfahler and Stamm in 1915, however its association with eosinophilic enteritis has been reported infrequently as entero-lithiasis in itself is rare [7]. Strictures of esophagus, pylorus and proximal small bowel in association with EG has been reported earlier. However, stricture formation in the terminal ileum due to EG with enterolithiasis is a rare occurrence. In the present case, the patient had ileal stricture due to EG with entero-lithiasis.

2. Presentation of case

A 55-year-old female presented with complaints of pain in abdomen in the periumbilical region, constipation and vomiting since 6 months. Pain was colicky in nature, intermittent, occurring once or twice a day, aggravated on taking food. She also complained of vomiting which was non-projectile, containing food particles and occurring once every 2–3 days. Patient also gave history of constipation, passing hard stools every 2–4 days with only partial relief with over-the-counter stool softeners. Patient had no medical comorbidities, allergic tendency or significant

Abbreviations: EG, Eosinophilic gastroenteritis; CECT, Contrast enhanced computed tomography.

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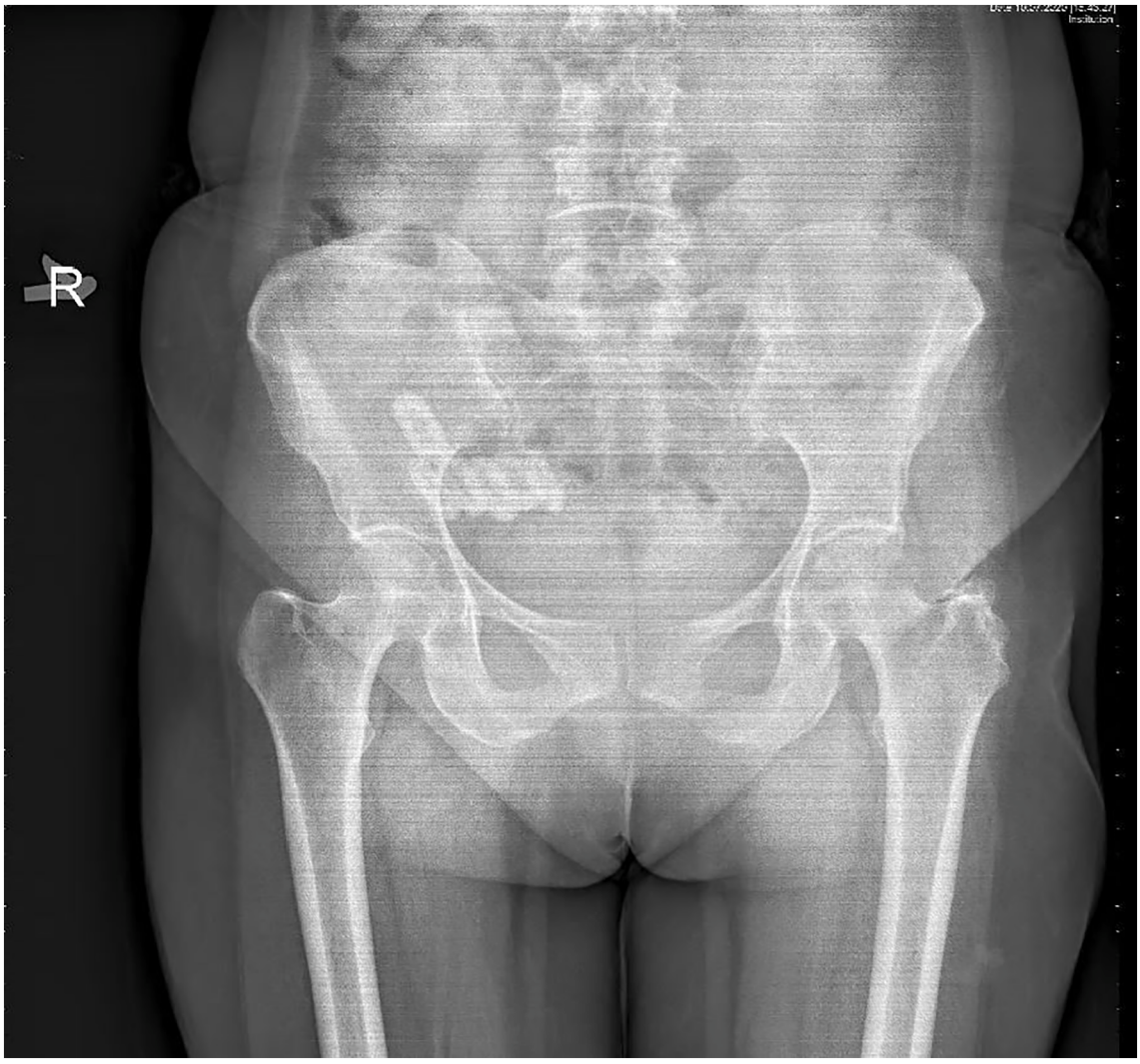

Fig. 1. Erect Abdominal Radiograph shoeing multiple well defined hyperdense structures (enteroliths) in right lower abdomen.

past surgical history. General examination and abdominal examination were unremarkable. Her complete blood count (CBC), renal and liver profile were unremarkable. Erect abdominal radiograph showed multiple well defined hyperdense structures in the lower part(Fig.1). Contrast enhanced computed tomography (CECT) scan with oral contrast showed wall thickening involving the terminal ileum, ileocecal valve and caecum. Ileal thickening was of 3.3 cm in length. Ileal stricture was seen with dilated proximal loops and enteroliths (Fig. 2A,B). Patient was worked up and planned for surgery in view of ileal stricture. Exploratory laparotomy was performed with a midline incision. An ileal stricture of approximate length of 10 cm was found at a distance of 100 cm from the Ileo-caecal junction with thickening of the mesentery. Decision was taken for resection of stricture segment with end-to-end anastomosis. A twenty-five centimetre length of ileal segment containing the stricture was resected(Fig. 3A). The segment contained seven hard, black stones of approximate size 3 × 1 cm each (Fig. 3B). It also contained a diverticulum on the mesenteric border which showed normal small bowel rugosity(Fig. 4). Histopathology of resected ileum showed eosinophilic enteritis and dense inflammatory infiltrate, predominantly containing eosinophils, lymphocytes and plasma cells in the mucosa, submucosa and muscularis propria (Fig. 5A,B). Submucosa was oedematous and showed lymphoplasmacytic infiltrates. Stone Analysis reported dense faecal matter with bile salts. Post-operative course in hospital was unremarkable with uneventful recovery. Follow-up of 9 months has shown her to be disease and symptom free.

3. Discussion

Eosinophilic gastroenteritis (EG) was first described by Kaijser in 1937 [3]. EG is a chronic inflammatory disease with poorly understood pathogenesis, characterized by variable degrees of eosinophilic infiltration within the gastrointestinal tract. It may mimic acute abdominal condition due to its wide spectrum of clinical presentation. EG occurs more commonly in third to fifth decades of life, though it might occur over a wide age range from infancy through the seventh decade [3,5]. Present case was a 55 years old female.

Eosinophilic gastrointestinal (GI) disorders may be primary or secondary. Primary eosinophilic GI disorders are the ones which occur in the absence of other known causes for eosinophilia (e.g. drug reactions, parasitic infections, and malignancy), selectively affect the GI tract with eosinophil-rich inflammation [7] as seen in present case. EG has a complex pathogenesis and is not well understood. EG can involve eosinophilic infiltration in any part of gastrointestinal tract. Although the mucosa normally has small amount of eosinophils as a host defence mechanism, but their presence in the deeper layers is uncommon. The criteria for the pathological diagnosis is the demonstration of eosinophilic infiltrate more than 20 per high power field [8]. This was detected by Desreumaux et al. in duodenal and colonic tissue in 90% of patients with EG [9]. Secondarily eosinophilic intestinal inflammation can occur in setup of inflammatory bowel disease, autoimmune diseases, reactions to medications, infections, hyper-eosinophilic syndrome and after solid

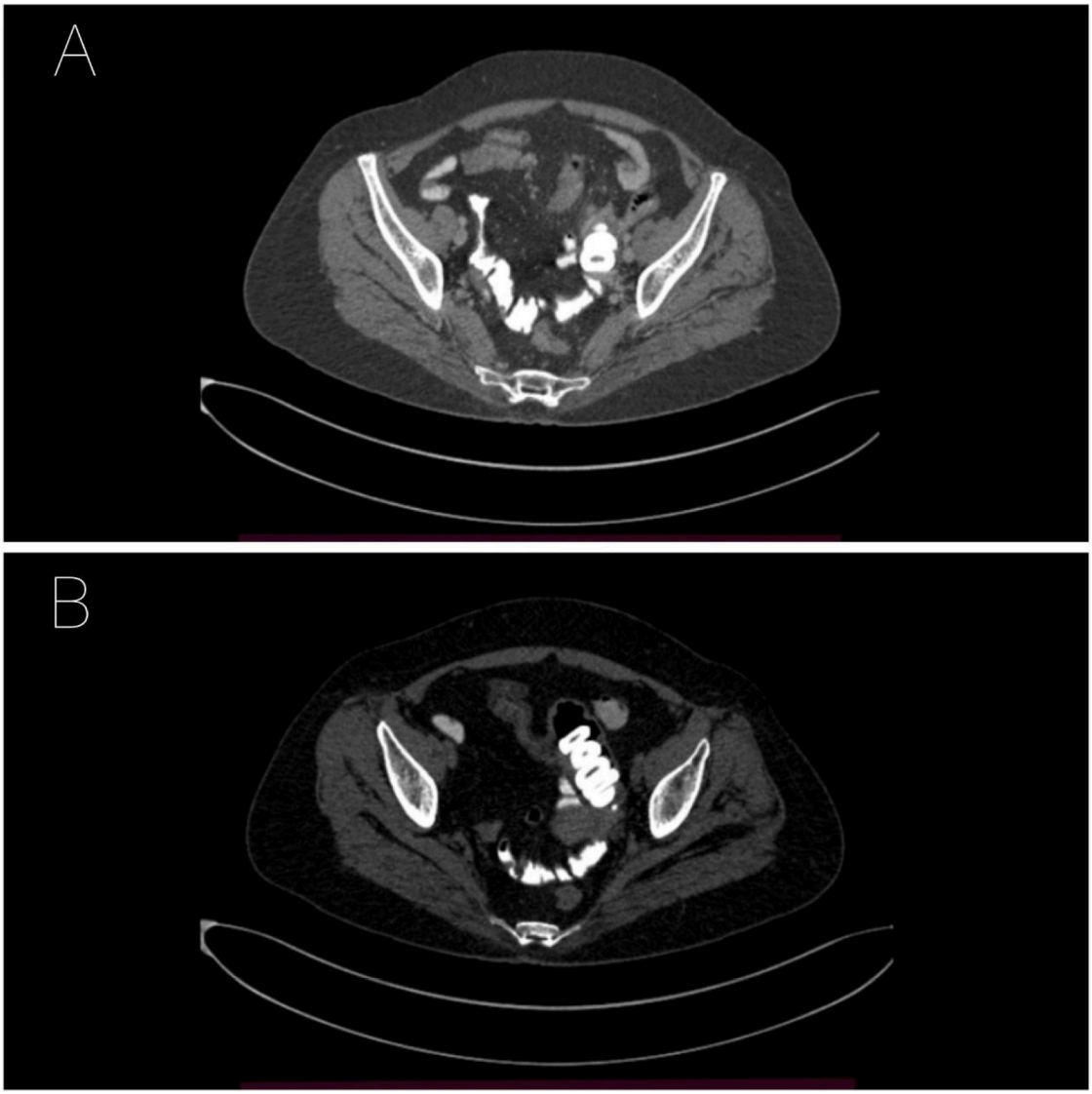

Fig. 2. A: CECT scan abdomen showing ileal stricture; B CECT abdomen showing enteroliths).

organ transplantation [10]. EG can be subclassified depending upon the segment of the gastrointestinal tract involved. Most cases involve the stomach and proximal small bowel, however, ileum was involved in present case. Talley et al. identified three main diagnostic criteria [11].

1. Non-specific gastrointestinal symptoms
2. Eosinophilic infiltration of one or more areas of the GIT
3. Exclusion of other causes for the intestinal eosinophilia

Present case fulfils all the above mentioned criteria. Klein classified the disease according to the predominance of eosinophilic infiltration in different layers of the intestinal wall [5] which include mucosal, muscular, and subserosal involvement. Patients present with wide spectrum of clinical presentation, depending upon the layer of involvement. Mucosal form is the commonest which presents with features of protein losing enteropathy, bleeding malabsorption or anaemia. Involvement of the muscularis layer leads to abdominal pain with nausea and vomiting with Entero-lithiasis was first described by Pfahler and Stamm in 1915. However, entero-lithiasis in association with eosinophilic enteritis has been reported in very few cases [7]. Hypomotility or stasis have been thought to be the main factors leading to enterolith formation, though other conditions might also be involved [12]. In present case, patient had ileal stricture resulting in prolonged stasis which could have contributed to the formation of enterolith and diverticulum.

Primary enteroliths are classified as true enteroliths and false enteroliths [13]. True enteroliths are formed by precipitation and deposition of substances from alimentary chime [13]. There are three main types of true enteroliths:(I) those consisting mainly of bile acids (II) those consisting mainly of calcium oxalate and (III)those consisting mainly of phosphate. Choleic acid is the main component of bile acid enteroliths [7]. False enteroliths results from clumping together and inspissation of intestinal content [12]. Secondary enteroliths are formed in the associated organs like gall bladder. Biochemical analysis of the stones recovered reported dense faecal matter with bile salts, which fits in the category of primary true enterolith. Enterolithiasis can be seen as typical dense rim with pale core in oval, round, or rectangular shadows "Coin-end-on" appearance of the shadows. Water-soluble contrast study combined with CECT can aid in diagnosis of sub-acute intestinal obstruction by enterolith [13]. CECT of present case showed coin end on appearance.

Imaging features of eosinophilic gastroenteritis include bowel wall thickening, layering of the bowel wall, diffuse mucosal fold thickening and luminal narrowing with or without intestinal obstruction. "Halo sign" and "Araneid limb-like" sign seen in inflammatory pathologies, helps in differentiating eosinophilic gastroenteritis from neoplastic

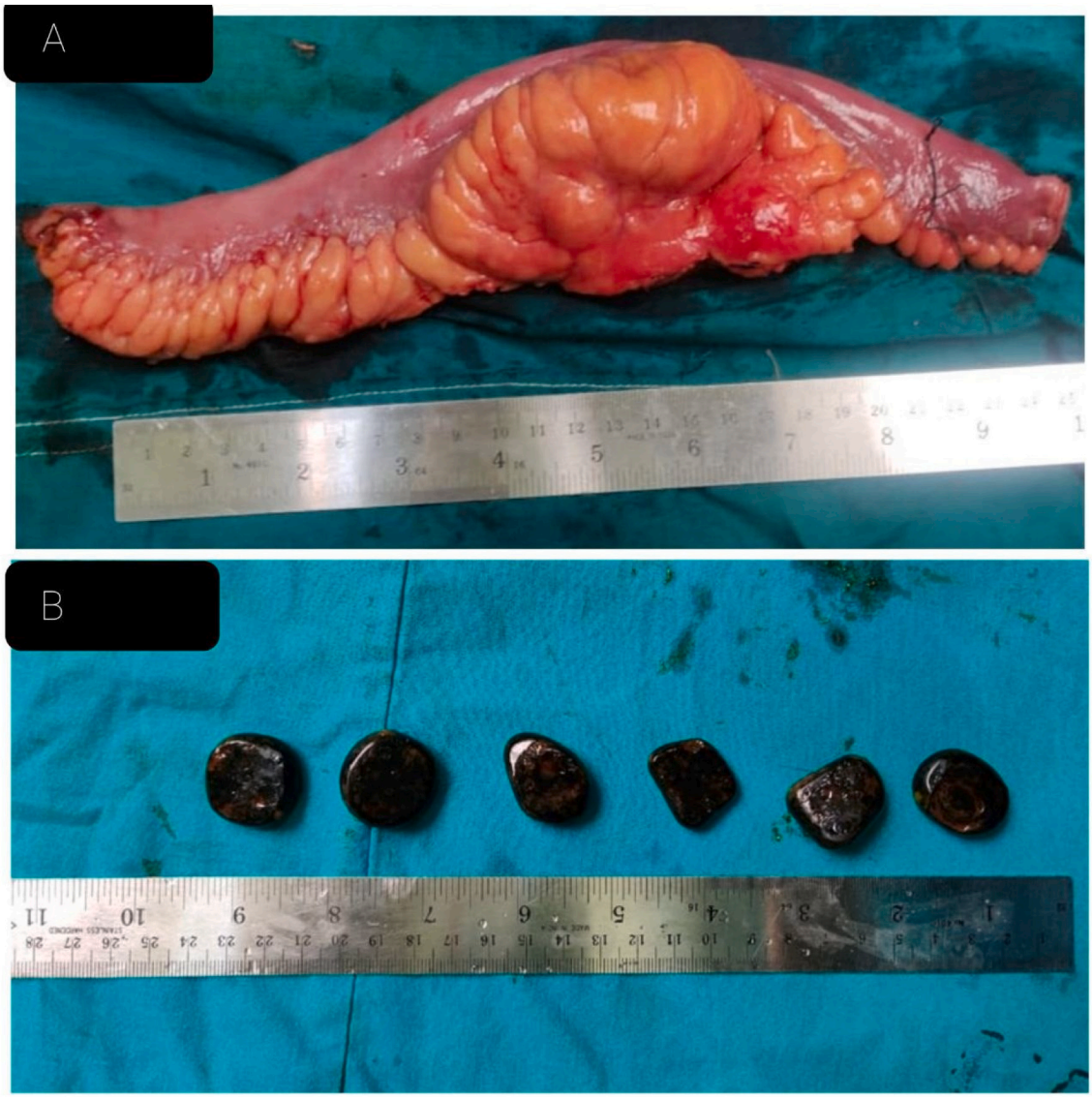

Fig. 3. A: Resected Ileal Segment; B: Stones in the resected ileal segment).

conditions, such as lymphoma and carcinoma [14]. CECT of present patient showed mild submucosal fatty infiltration with wall thickening involving terminal ileum, ileo-caecal valve and caecum. Endoscopic biopsy can aid in diagnosis, however in patients with muscular or serosal subtype or both, biopsies might be normal.

As patients often present with a wide variety of signs and symptoms with non-specific laboratory and radiological findings, this condition is often mistaken for similar disorders during the initial workup period, ultimately leading to a delay in diagnosis. EG once diagnosed has been treated with steroids, sodium chromoglycate, sodium montelukast and even immunosuppressive drugs like azathioprine [15,16]. However, once stricture is formed with symptoms and signs of intestinal obstruction, surgery is the mainstay of treatment. The definitive diagnosis is made after histological examination of the surgical specimen. Present case had stricture with symptoms of subacute intestinal obstruction, hence surgery was the treatment of choice with histopathology of the resected specimen confirming the diagnosis.

4. Conclusion

Eosinophilic Gastroenteritis (EG) due to its rare occurrence, overlapping clinical features, laboratory and radiological findings may mimic other GI disorders resulting in misdiagnosis or late diagnosis. Surgery is the ideal treatment recommended in stricture causing intestinal obstruction or perforation. A high index of suspicion is required to diagnose EG. It should be considered as one of the differentials in chronic abdominal pain with varied presentation.

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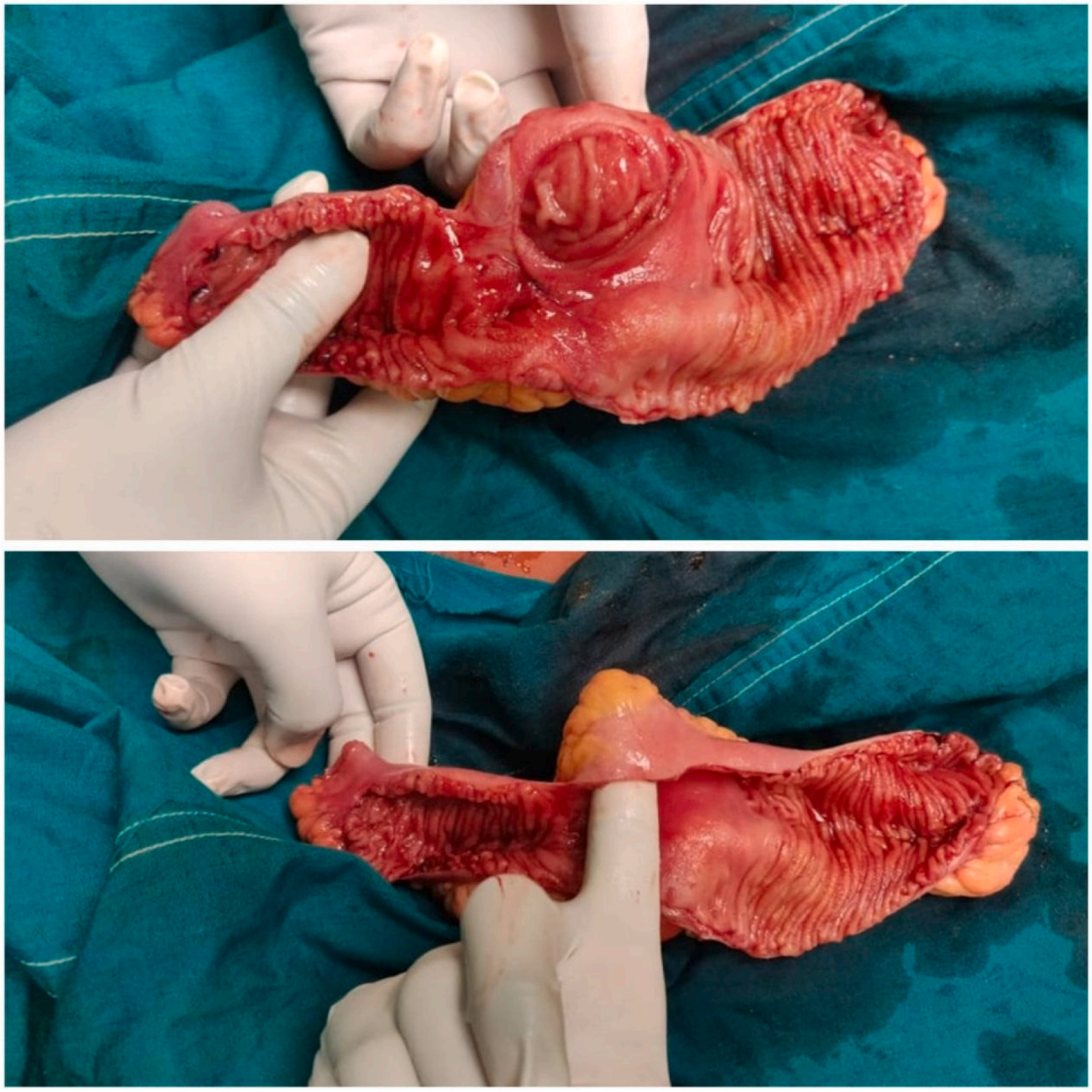

Fig. 4. Diverticulum on the mesenteric border.

Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Research registration

Not applicable.

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CRediT authorship contribution statement

Dr. Girish D. Bakhshi: Study design, Performed the surgery.
Dr. Md Furqan Abdul Waheed: Assisted for surgery, Data collection, Writing the paper and is corresponding author.
Dr. Zarin Rangwala: Assisted for surgery, Data collection.
Dr. Owais Ahmed Patel: Assisted for surgery, Data collection.
Dr. Aishwarya Mohan: Assisted for surgery, Data collection.
Dr. Urvashi Jain: Assisted for surgery, Data collection.

Declaration of competing interest

None.

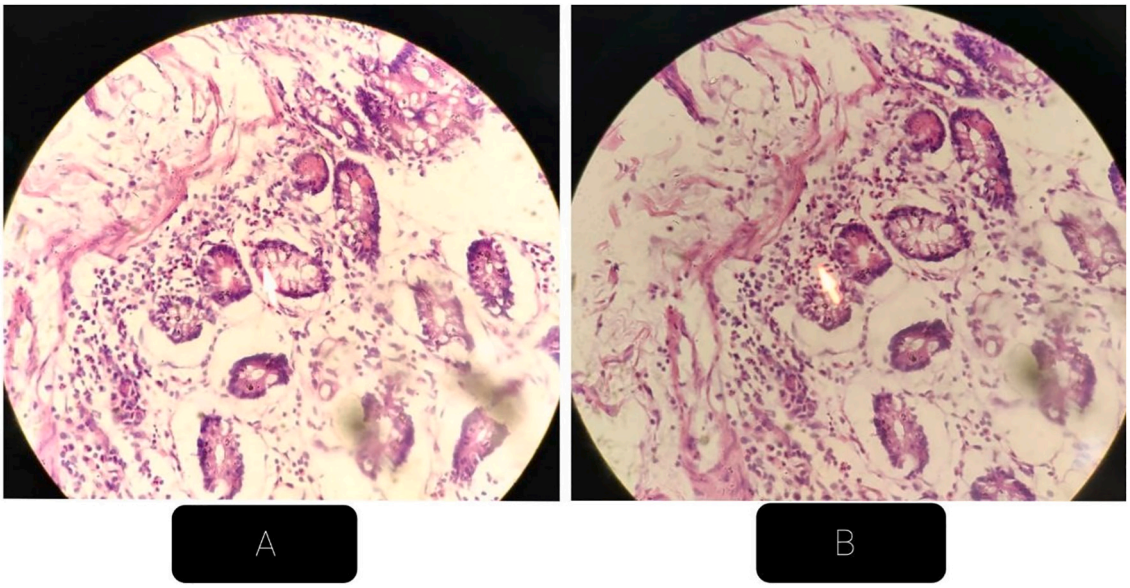

Fig. 5. A: White Arrow showing eosinophilic infiltration in the mucosa; B: Arrow showing eosinophilic infiltration in the submucosa {Hematoxylin and Eosin stain, 10×}).

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