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Eosinophilic Enteritis Presenting in Emergency Like as Small Bowel Obstruction: A Case Report and Literature Review

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Abbreviations:
EE: Eosinophilic enteritis;
EGIDs: eosinophilic gastrointestinal disorders;
CECT: contrast-enhanced computed tomography;
GI: gastrointestinal.
EGID: Eosinophilic gastrointestinal disease.

ABSTRACT

Introduction: Eosinophilic enteritis (EE) is an uncommon condition which belongs to primary eosinophilic gastrointestinal disorders (EGIDs). The pathogenesis and etiology of EGIDs remain uncertain. Nowadays there are no gold-standard diagnostic criteria for EGIDs. The first-line treatment for EGIDs is glucocorticoid administration. Surgery may be recommended when a definitive diagnosis of EGIDs cannot be made and it is mandatory when complications occur.
Case report: A 72-year-old Caucasian female presented to the Emergency Department with a two-day history of diffuse abdominal pain, nausea, vomiting and inability to pass faeces and gas. Abdominal examination showed abdominal distention, diffuse abdominal pain without obvious muscle guarding and rebound tenderness. Blood exams revealed high levels of C-reactive protein and neutrophilic leukocytosis without eosinophilia. Abdominal contrast-enhanced computed tomography (CECT) revealed a circumferential small-bowel wall thickening causing small bowel obstruction. The patient underwent exploratory laparotomy: a focal double stenosis in the ileum was found and a segmental resection of the ileum was performed. The postoperative course of the patient was unremarkable.
Conclusion: EE is a rare disease of unknown etiology and represents an uncommon cause of small bowel obstruction. A strong clinical suspicion and awareness of this disease are essential to obtain a correct diagnosis.
Key words: eosinophils, eosinophilic enteritis, intestinal obstruction, endoscopy, surgery.

INTRODUCTION

Eosinophilic enteritis (EE) is a rare disease which belongs to primary eosinophilic gastrointestinal disorders (EGIDs). Primary EGIDs are characterized by the accumulation of eosinophils in the gastrointestinal tract and include eosinophilic esophagitis, gastritis, enteritis and colitis (1). Primary EGIDs are strongly associated with atopy and allergy in the absence of other causes of eosinophilic infiltration (secondary EGIDs) like as inflammatory bowel disease, parasitic infections, drug reactions and malignancies (1,2). With approximately

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300 reports found in the literature prevalence of primary EGIDs remains low, however it has increased in the last years with a female gender and Caucasian race predilection (3,4). Although the pathophysiology remains uncertain, EE is characterized by chronic intermittent period of inflammation with eosinophilic infiltration of the small bowel wall to a variable depth. EE can present in various ways depending on the site and extent of eosinophilic inflammation (5). We present a case of EE that underwent emergency laparotomy for acute small bowel occlusion.

CASE REPORT

A 72-year-old Caucasian female presented to the Emergency Department with a two-day history of diffuse abdominal pain, nausea, vomiting and inability to pass faeces and gas; vital signs were normal. Her past medical history included arterial hypertension, dyslipidemia. The patient was on hypertensive medications for ten years and denied allergic disease and food sensitivity. Abdominal examination showed abdominal distention, diffuse abdominal pain on superficial and deep palpation without obvious muscle guarding and rebound tenderness, hypoactive bowel sound. Blood exams reported high levels of C-reactive protein (90.5 mg/L) and a white cell count of 16.700 103/µL with 85.9% neutrophils and 1% eosinophils. The patient was initially managed with fluids, intravenous broad-spectrum antibiotics and bowel rest. After a plain abdominal x-ray showing intestinal air-fluid levels (*fig. 1*), the patient was evaluated by abdominal contrast-enhanced computed tomography (CECT) which revealed at level of the medium ileum a circumferential small-bowel wall thickening causing small bowel obstruction and the presence of abdominal and pelvic free fluid (*fig. 2 a,b,c*). The patient was taken emergently to the operating theatre for exploratory laparotomy. Abdominal exploration revealed a double focal stricture in the ileum about 30 cm and 130 cm proximal to the ileocecal valve (*fig. 3 a,b,c*) and a non complicated Meckel's diverticulum between the double intestinal focal stricture. The intestinal narrowed lumen in the indurated area was believed to have been the cause of intestinal obstruction. A segmental resection of the ileum bearing the double focal stricture and the Meckel's diverticulum with latero-lateral mechanical ileo-ileal anastomosis was performed. After washing and aspiration of peritoneal cavity a laminar peri-anastomotic drain was placed. Patient was given an IV

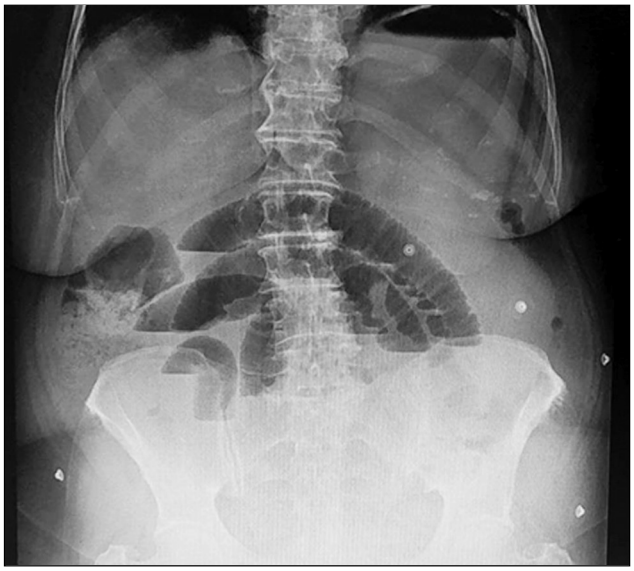

Figure 1 - Plain abdominal x-ray showing intestinal air-fluid levels.

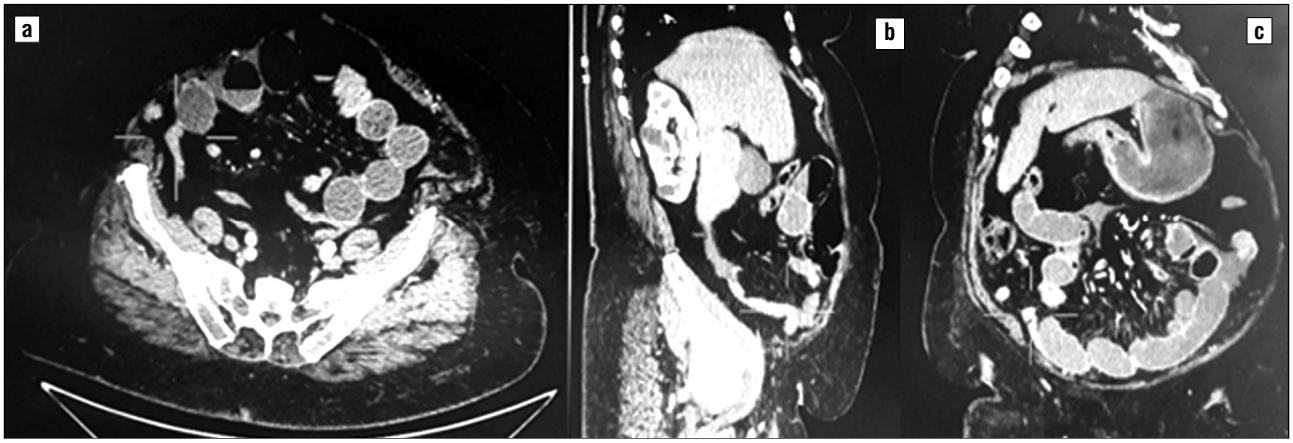

Figure 2 - (a,b,): Abdominal CECT showing a circumferential small-bowel wall thickening (yellow signal) causing intestinal obstruction. A transverse view, B sagittal view, C coronal view.

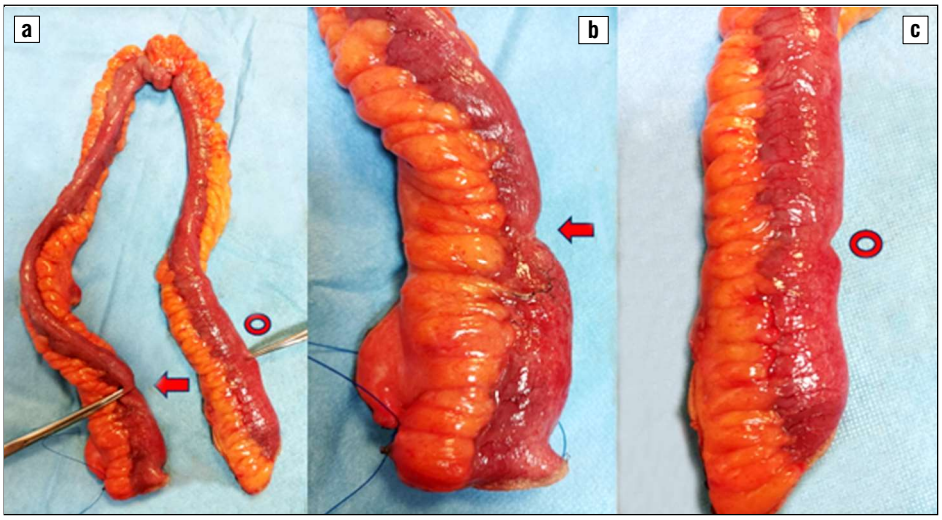

Figure 3 - (a): the surgical specimen consisted of the distal ileum bearing the double focal stricture about 30 cm (red arrow) and 130 cm (red circle) proximal to the ileocecal valve and the Meckel's diverticulum. (b): enlarged photo of the focal ileal stricture about 30 cm proximal to the ileocecal valve (red arrow). (c) enlarged photo of the focal ileal stricture about 130 cm proximal to the ileocecal valve (red circle).

injection of Amoxicillin/Clavulanate 2 gr twice daily and Metronidazole 500 mg thrice daily for five days. The postoperative course was uneventful, the patient was discharged on the 6th postoperative day, after removal of the abdominal drain, in a stable condition. Histopathological analysis of the surgical specimen revealed in the double focal intestinal stricture a transmural infiltration by inflammatory cells (mainly eosinophils) (*fig. 4 a,b,c,d*) and EE was diagnosed. No evidence of parasites, granulomas, malignancy, vasculitis or embolism was identified in the bowel wall or in the mesenteric vessels present in the surgical specimen. Postoperatively the serum IgE concentration remained normal and repeated examination of the patient's stool never revealed ova or parasites. During the follow-up period of 1 year the patient was completely free of recurrence. This clinical case is particular because EE represents a very rare cause of intestinal obstruction diagnosed postoperatively on histological examination.

DISCUSSIONS

Eosinophils are constituents of the gastrointestinal (GI) mucosa where they likely have a role in immune surveillance. However, when eosinophils are found in higher numbers, disease pathology may result, especially following eosinophil activation with resulting proinflammatory effects of their granule proteins. Eosinophilic gastrointestinal disease (EGID) is a rare inflammatory disease that presents with variable degrees of infiltration of eosinophils within the GI tract, first described by Kaijser in 1937 (6). Due to its rarity and heterogeneity, its exact prevalence and risk factors are difficult to classify. The recent prevalence of EGID ranges from 5 to 8 per 100.000 of the general

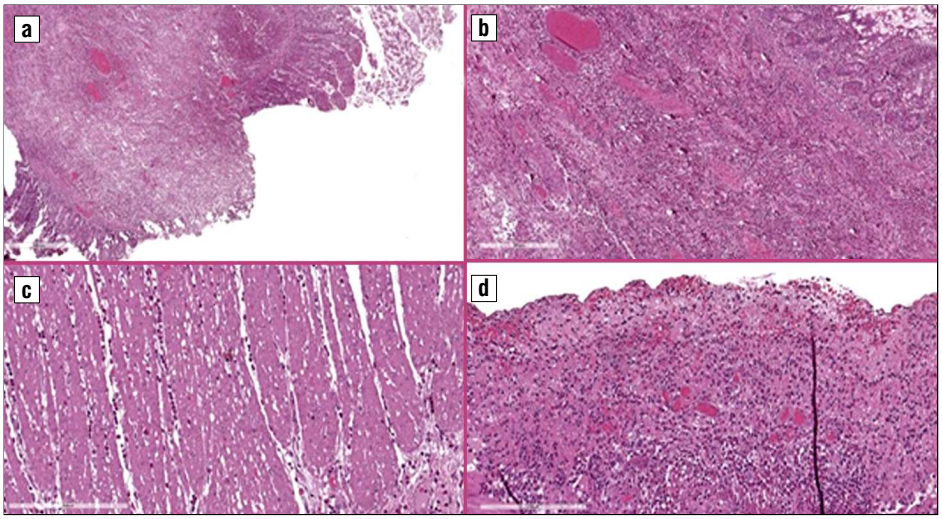

Figure. 4 - Photomicrographs of the resected ileum revealed transmural infiltration by eosinophils and ulceration of the mucosa and serosa. (a): ulceration of the mucosa. (b): submucosal eosinophilic infiltration. (c): eosinophilic infiltration in the muscular layer. (d): inflammation of the subserosa and serosa, ulceration of the serosa. Haematoxylin and eosin: original magnification x 20 (A), x 40 (B), x 200 (C), x 40 (D).

population (3). EGID can affect any age group with a female predominance (5); the median age of patients ranges from 30 to 50 years old. The pathogenesis and etiology of EGID remain unclear (2) although several reports have hypothesized a role of allergies to food and other allergens (7). Our patient had no history of allergies or food sensitivity. EGID can be sub-classified either according to the segments of the GI tract affected by the process or according to the depth of the eosinophilic infiltration. According to the segments of the GI tract affected, EGID can be divided into four subtypes: eosinophilic esophagitis, gastritis, enteritis and colitis. EGID can affect any portion of the gastrointestinal tract, although the stomach and duodenum are the most common. According to the depth of the eosinophilic infiltration, three main patterns of EGID can be delineated: predominant mucosal disease (type I), predominant muscle layer disease (type II), predominant subserosal disease (type III) (8). Currently there are no gold-standard diagnostic criteria for EGID. The widely used diagnostic approach is composed of three criteria: presence of GI symptoms, endoscopic biopsies showing histological presence of eosinophilic infiltration within the GI tract and extensive exclusion of other differential diagnoses of peripheral and/or tissue eosinophilia (9). The clinical presentation of EGID depends on the layer depth and the location of the affected tissue. The mucosal form (the most common subtype) is characterized by vomiting, abdominal pain, diarrhea, blood loss in stool, iron-deficiency anemia, malabsorption and protein losing enteropathy. The muscularis form is characterized by infiltration of eosinophils predominantly in the muscle layer and is more associated with bowel-wall thickening, intussusceptions, gastric outlet obstruction and intestinal obstruction as in our case report. The subserosal form is associated with the presence of eosinophilic ascites, peritonitis and GI perforation (10). Transmural disease, as in our case, presents with intestinal wall thickening and features of intestinal obstruction. The most common endoscopic finding of EGID is a normal appearance (62%), less common findings are erythema, ulceration, nodularity and mucosal friability (11). GI tract biopsy is a critical factor in making the diagnosis of EGID regardless of endoscopic appearance: multiple random biopsies are warranted per site (at least 5-6 biopsies). In case of muscular or subserosal disease, endoscopic biopsies often fail and surgically obtained full-thickness or fine needle aspiration biopsies are necessary to obtain a histological diagnosis. Wireless capsule endoscopy may also play a diagnostic rolebut tissue sampling is required (12). Although there is no consensus regarding the eosinophilic cut-off for diagnosis, several studies have suggested histologic criteria for the diagnosis of EGID including ≥ 30 eosinophils/HPF (high-power field) in at least 5 HPF of the stomach for eosinophilic gastritis and > 20 eosinophils/HPF in the duodenum or at least 56 eosinophils/HPF in the ileum for eosinophilic gastroenteritis. For eosinophilic colitis ≥ 100 eosinophils /HPF in the cecum/ascending colon, ≥ 84 eosinophils /HPF in the transverse/descending colon, and/or at least 64 eosinophils/HPF in the rectosigmoid colon may indicate disease (13). In our case report the histopathological examination of the surgical specimen revealed eosinophilic infiltrate throughout the entire thickness of ileal wall >60 eosinophils/HPF. The differential diagnoses of peripheral and tissue eosinophilia is broad including systemic disorders, food allergy, drug induced, oncologic, inflammatory and infectious causes (5). The primary differential diagnoses to consider are intestinal parasites, ulcerative colitis, Crohn's disease, intestinal malignant lymphoma, neoplasm, tuberculosis, collagenous colitis, scirrhous gastric cancer, celiac disease, protein-losing gastroenteropathy and hypereosinophilic syndrome (14). Multiple studies show that about 70% of EGID patients present with peripheral eosinophilia (>500cells/mL) (15) which represents one of the signs of disease severity, however it was not present in our patient. An elevated total IgE concentration is observed in more than 70% of affected patients, though their diagnostic role remains unclear (16). Radiological exams (abdominal CECT or ultrasonography) frequently show ascites and segmental wall thickening of the involved gastrointestinal tract (17), however these radiological findings are nonspecific. In our case report abdominal CECT revealed a single small-bowel wall thickening causing intestinal obstruction but intra-operatively a double focal small bowel stricture was observed. No prospective randomized controlled study regarding the treatment of EGID has been conducted mainly due to its low prevalence. Dietary restrictions, antiallergic drugs, proton pump inhibitors, gluco-corticoids and molecular targeted drugs are used for the treatment of EGID. Based on limited case reports and clinical experience, first-line treatment for EGID remains glucocorticoid administration. Surgery may be recommended when a definitive diagnosis of EGID cannot be made and is mandatory when complications (GI obstruction, intussusception or perforation) occur (18,19). Mortality is rare although the natural

history and prognosis of EGID are unpredictable: the risk of relapse or recurrence, even after surgery, is common and closely associated with high eosinophil counts, so long-term follow-up is required.

CONCLUSIONS

EE is a rare disease of unknown etiology and represents an uncommon cause of small bowel obstruction. Currently there are no gold-standard diagnostic criteria for EE. A strong clinical suspicion and awareness of this disease are essential to obtain a correct diagnosis. Until now corticosteroid treatment remains the main therapy. Surgical managementis indicated in the complications of EE.

Conflict of interest

The authors declare that they have no conflict of interest.

Ethics approval

The study case was carried out in accordance with the Declaration of Helsinki on experimentation with huma subjects.

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