A Rare Case of Eosinophilic Colitis

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Abstract

Background: Eosinophilic Colitis (EC) is the least well-understood subgroup of the Primary Eosinophilic Gastrointestinal Diseases (EGIDs), a spectrum of disorders characterized by pathologic eosinophilic infiltration along the gastrointestinal tract, in the absence of other causes of eosinophilia. It is currently regarded as a diagnosis of exclusion.

Case Presentation: This is a case report of EC, where a 47-year-old gentleman presented with chronic diarrhoea, mild abdominal pain, weight loss and intermittent right forearm swelling. Significant eosinophilia (7.63%) was identified and colonic biopsy demonstrated focal superficial active inflammation with markedly increased eosinophilic infiltration in the lamina propria. The work up for hyper-eosinophilia was otherwise negative. Once oral corticosteroids were initiated, his symptoms and eosinophil count improved.

Conclusion: The scarcity of the reporting for EC could be due to its under-diagnosis. We suggest that all cases of chronic diarrhoea should be offered colonoscopy with multiple biopsies to rule out the possibility of EC. A number of immunomodulatory agents and biologic agents are thought to hold promise for the treatment of EC. Due to its undefined pathophysiology, diagnostic thresholds and codified treatment guidelines, further studies involving large sample sizes of patients are required to understand its immunopathophysiology, in order to define optimum management strategies.

Keywords: Eosinophilic; Eosinophilic gastrointestinal diseases; Corticosteroids

Introduction

Primary Eosinophilic Gastrointestinal Disorders (EGIDs) represent a spectrum of inflammatory gastrointestinal disorders in which eosinophils intensely infiltrate the gastrointestinal tract segmentally or diffusely, without evidence of other causes for eosinophilia. EGID can be sub-grouped into: Eosinophilic Oesophagitis (EO), Eosinophilic Gastritis (EG), Eosinophilic Gastronenteritis (EGE), Eosinophilic Enteritis (EE) and Eosinophilic Colitis (EC). Amongst EGIDs, there is relatively little information concerning EC, its rarest form [1].

The actual prevalence of primary EGID is largely unknown in the presence of secondary causes of eosinophilic inflammation such as parasitic and helminth infections, IgE-mediated allergy reaction, drug side effect, autoimmune disorders, hypereosinophilic syndrome, gastro-oesophageal reflux disease and inflammatory bowel disease [1]. Rothenberg et al. report that EC appears to have a bimodal distribution, mainly affecting neonates and young patients with no gender predominance [1].

Although the aetiology of EC remains unclear, genetic and allergenic components may play a role.

Some studies report that 16% of patients with EGID have a positive family history and 80% have a co-existing atopic disease [2,3]. Although cow’s milk and soy protein intolerance have been implicated in the infantile form of EC, potential food association in the adult form of the disease...
is still undefined [4]. The pathophysiology of food hypersensitivity in EC may be attributed to the pathogenic role of IgE, where mast cell accumulation is visualized in the colonic biopsy [5]. However, in adults, EC is mostly non-IgE associated, involving a CD4(+) Th2 lymphocyte-mediated mechanism [6]. In EC, the endoscopic changes are non-specific. Currently, there is no consensus on the histological criteria for the diagnosis of EC because there are no specific limits for normality. Most experts regard twenty eosinophils per high-power field as the diagnostic threshold for dense colonic eosinophilic infiltration [4]. Other supportive evidence can be obtained from allergic skin testing via Skin Prick Test (SPTs) and Radioallergosorbent Test (RASTs) where a negative test is required to exclude IgE-mediated food allergy [1]. Clinical presentations in EC vary according to which colonic layers are predominantly affected; but generally, include abdominal pain, diarrhea and weight loss [1]. There is a scarcity of case reports of EC in the West. Here is a case of EC in a 47-year-old Caucasian gentleman, who responded well to a tapering course of prednisolone. Our report will alert clinicians to the presentation of EC and its subsequent treatment with steroid therapy, after careful evaluation has ruled out alternative diagnoses.

**Case Presentation**

A 47-year-old gentleman was admitted with eighteen months of chronic diarrhea and four months of weight loss of five kilograms and intermittent right forearm swelling. Diarrheal episodes were loose, occurring three to four times a day without any mucus or blood. He complained of dull, steady and mild epigastric pain which was non-radiating, without any aggravating or relieving factors. There was no history of jaundice, change in appetite, dysphagia, heartburn, fever, rash, wheezing or B symptoms. He visited his GP on few occasions, who attributed his symptoms to food poisoning or IBS.

He had suffered from urticaria and allergic rhinitis for ten years. Other past medical history included: Heart failure due to non-ischemic cardiomyopathy in January 2019, type 2-diabetes mellitus with previous diabetic retinopathy and a decade of Essential Hypertension. The patient had no known pre-existing food or drug allergies. He denied recent new drug usage such as herbal medicines and NSAIDs. He was a non-smoker and a teetotaler, with no recent foreign travel. Physical examination was unremarkable. His investigations showed; hemoglobin 120 g/dL; WBC 12.76 (neutrophils 3.55, eosinophils 7.63; Lymphocytes 1) and platelets 329. Total IgE 4774 (0-81). His stool and parasitic serological investigations and bacterial stool culture were negative. Liver, renal and thyroid function tests were normal but biopsies revealed preserved architecture with mild distortion of glands, focal superficial active inflammation and large numbers of eosinophils in the lamina propria (Figure 1). There was no evidence of colitis, dysplasia or malignancy.

On this admission, flexible sigmoidoscopy was performed and multiple biopsies were obtained. Macroscopic appearances were normal but biopsies revealed preserved architecture with mild distortion of glands, focal superficial active inflammation and large numbers of eosinophils in the lamina propria (Figure 1). There was no evidence of colitis, dysplasia or malignancy.

**Results and Discussion**

A diagnosis of EC was made and the patient was started on a tapering course of oral prednisolone. Upon follow-up review, the patient had complete resolution of his bowel movement, with fully formed stool and eosinophil count fell to 0.1 (Figure 1). The pathophysiological mechanisms for EC remain understudied. EC is mainly a diagnosis of exclusion after secondary causes have been thoroughly investigated, with three hallmarks: Presence of peripheral eosinophilia, eosinophilic infiltration of the gastrointestinal tract and functional abnormalities [7,8]. Our patient fulfilled the diagnostic criteria with the presentation of chronic diarrhea and peripheral eosinophilia, combined with histological findings of dense eosinophilic colonic infiltration. His diarrheal symptoms and eosinophil count resolved once a tapering dose of steroids were initiated and clinical remission was evident on discharge. With lack of current guidelines or consensus in the management of EC, treatment is mainly based on case reports and small case series. It is empiric and based on the severity of symptoms; largely involving dietary exclusions of potential food triggers or medical therapy to control symptoms and bowel inflammation. (Surgery is only indicated in intestinal obstruction or perforation.) Young children with eosinophilic proctitis may achieve clinical remission on elimination of an oligoantigenic and amino acid-based diet [3]. However, in adult and adolescent EC where natural history shows chronicity with a relapsing and remitting pattern, corticosteroids are generally recommended, for their potential for inhibiting eosinophilic proliferation [3,4]. Despite the absence of randomized controlled trials to date on the efficacy of steroids in EC, the majority of cases in practice (such as ours) result in clinical remission within two weeks of treatment with 20 mg to 40 mg prednisolone [4].

Budesonide has been reported to achieve maintenance of remission for up to two years, especially where the right colon and ileum are affected [9]. Because patients have been reported to relapse with steroid withdrawal, there has been more recent emphasis on steroid-sparing treatment. Immunomodulatory agents such as Azathioprine have been shown to be effective, especially in steroid-dependent disease [1]. Mast cell stabilisers such as Cromolyn Sodium and leukotriene receptor antagonists such as Montelukast have also shown promise in some studies [10]. Finally, there is an emerging focus on the clinical development of novel biologic agents including humanised anti-IL5 antibodies for the treatment of EC [10].

**Conclusion**

EC remains a rare disorder, with undefined pathophysiology.
diagnostic thresholds and codified treatment guidelines. Being a diagnosis of exclusion, a careful systemic evaluation is required by clinicians for its recognition. Further studies involving large sample sizes of patients are required to understand its immunopathophysiology, in order to define optimum management strategies.

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Author’s Contribution

VS was the major contributor in writing the manuscript, obtaining investigation results from the patient’s clinical notes and obtaining the image from Histopathology. NS contributed to finding a suitable article for publication. NC contributed to the final manuscript and finding a suitable article for publication. GA supervised the entire process. All authors read and approved the manuscript.

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