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Case Report

Asymptomatic Eosinophilic Esophagitis in apatient with very early-onset ulcerative colitis

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Eosinophilic esophagitis (EoE) is a chronic inflammatory disorder of the esophagus characterized by symptoms of dysphagia or GERD-like symptoms, specific endoscopic findings and eosinophilic infiltrates. We report a case of asymptomatic eosinophilicesophagitis in a 3 years old child who is known to have very early-onset ulcerative colitis at 8 months and diagnosed at 18 months of age.

Keywords: Eosinophilic Esophagitis, ulcerative colitis,

INTRODUCTION

Eosinophilicesophagitis (EoE) is a chronic disease characterized by chronic esophageal mucosal inflammation with denseinfiltration of eosinophils (Yuji et al., 2011; Furuta et al., 2007). EoE is frequently diagnosed in the last few decades in children and adults. EoE has been reported in association with other disorders like celiac disease, food allergy, and Crohn's disease (Furuta et al., 2007). In this paper, we report a young child with a symptomatic EoE and very early-onset ulcerative colitis (UC) and discuss the significance of this association.

The case

The child presented at the age of 18 months with a one week history of bloody diarrhea (5 to 6 times per day), abdominal pain, poor weight gain and fair appetite since the age of 8 months. He had no history of vomiting, dysphagia or chocking. He received breast feeding till 4 month of age then weaned to regular diet for age. His vaccination was up to date and he hadno history of food allergy. Past medical history was unremarkable except for eczema and mild developmental delay. The parents are first degree cousins. The father and two sisters are healthy but the mother has ulcerative colitis in remission.

Physical examination on first admission showed a well looking child with mild pallor but no jaundice, cyanoses, ordimorphic features. Pulse 110/min, respiratory rate 20/min, SPO2 98% in room air, BP 90/60

Weight was 13 Kilograms and height was 93 centimeters (both at the 25th percentile). The skin, musculoskeletal system, chest, cardiovascular examination were normal. The abdomen was not distended, not tender and no organomegaly was found. He had perianal rash and abscess but no fissures or fistulas were seen. Investigations included: Hb% 99gm/l, WBC 8.7 X10^3/l differential eosinophil 0.7%, nutrophil 44%, lymphocytes 42%. Monocytes 12%, platelets 500X10^6 , ESR 59mm/hr, LFT, U&E were normal, albumin 23gm/l, celiac markers were negative.

Complete immunologic investigations showed Immunoglobulins IgE 310 ku/l(normal up to 100), IgG 7.87gm/l (5 - 13), IgA 0.44gm/l (0.14 – 1.08), IgM0.697gm/l (0.43 – 2.39). lymphocytes markers and blastogenesis were normal.

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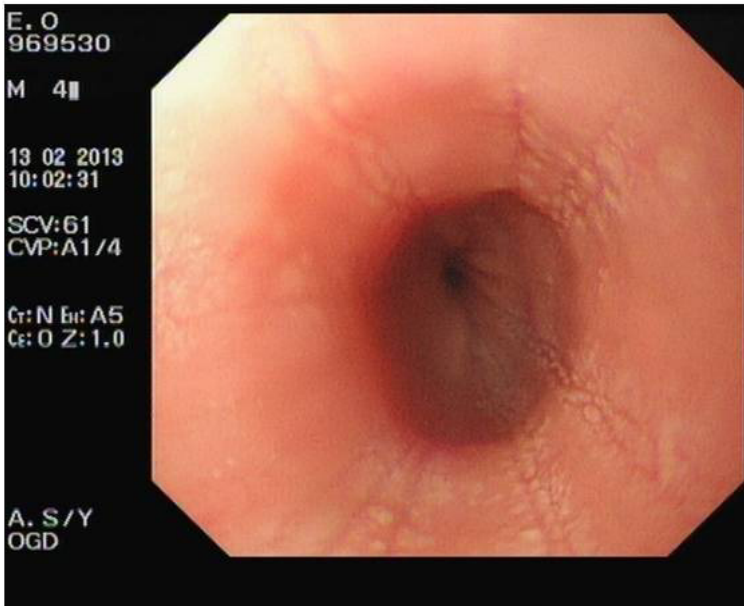

Figure 1. EGD: furrowing of the esophageal mucosa white patches

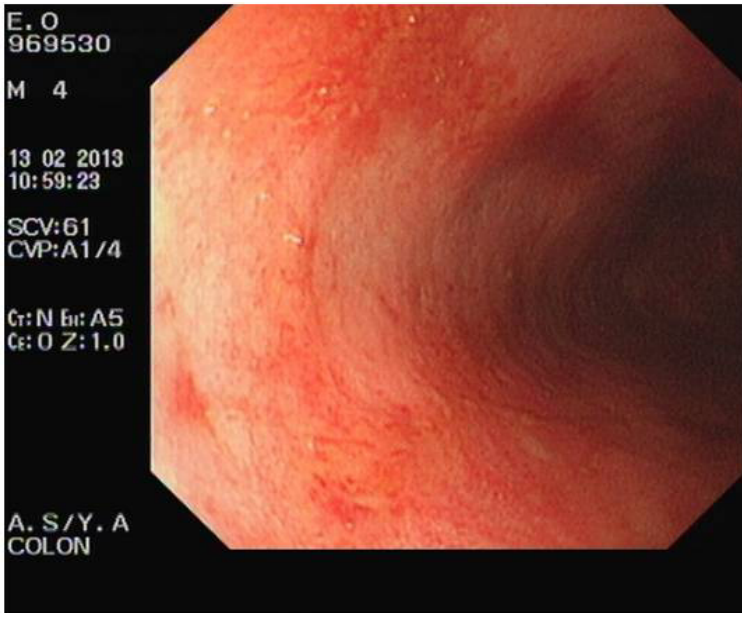

Figure 2. Colonoscopy: multiple ulcerations involving the whole colon more severe in the rectosigmoid

The first colonoscopy performed at presentation (18 months of age) showed features of distal colitis and the histopathology showed active chronic inflammation, distorted crypts architecture and abscesses, consistent with inflammatory bowel diseases most probably UC.

He was treated initially with sulphasalazine and prednisolone with good response. Prednisolone was tapered and discontenued and continued on sulfaslazine. He had few mild relapses on sulfasalazine requiring short courses of prednisolone. During the last relapse in the form of bloody diarrhea and fever, reevaluation in the form of esophago-gastro-duodenoscopy (EGD) and ileocolonscopy were performed. EGD showed furrowing of the esophageal mucosa with white patches (Figure 1) consistent with EoE, and ileocolonoscopy showed multiple ulcerations involving the whole colon more severe in the rectosigmoid (Figure 2). The terminal ileum was normal. Histopathology of the esophageal mucosa

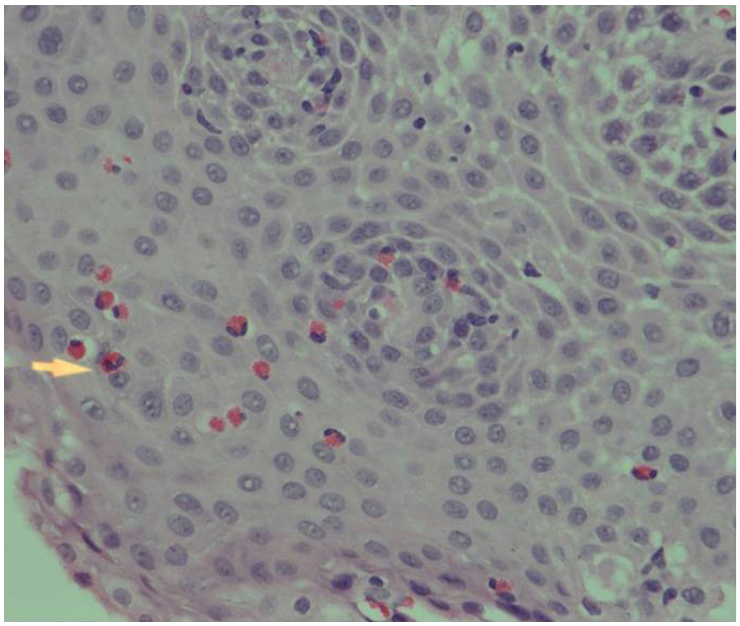

Figure 3. Esophageal biopsy showing numerous inttraepthelial eosinophils consistent with eosinophilic esophagitis. H/E stain x400.

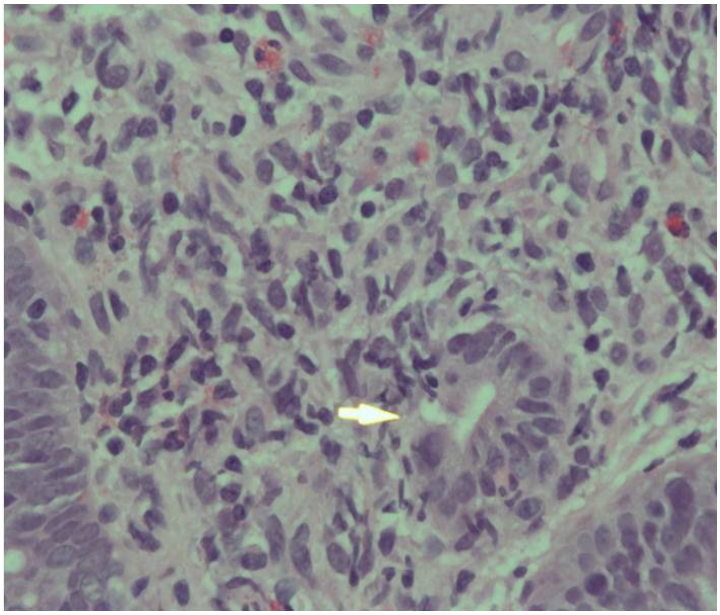

Figure 4. Colonic biopsy showing marked acute on chronic inflammation in the lamina propria with evidence of goblet cells depletion and early crypt abcess formation. H/E stain x400

showed increased eosinophilic infiltrates (>15/HPF) consistent with EoE (Figure 3). Histopathology of terminal ileum was normal, but biopsies of the cecum, ascending colon, transverse colon, descendingcolon and sigmoid colon, showed moderate chronic active colitis with architectural distortion cryptitis and crypt abscesses and no significant eosinophilic infiltration was demonstrated. These findings are consistent with active chronic inflammatory bowel disease (Figure 4).

He underwent allergy testing RAST test was done for 41 food allergens including milk, egg, fish and bean nut all came negative and couldn't identified specific offending food allergies. He was given singulair together with IBD treatment

DISCUSSION

EoE is increasingly recognized as a distinctcondition with esophageal symptoms, being a common cause of dysphagia, food bolusimpaction, and retrosternal chest pain. The association with IBD has been reported mainly from Western countries with Crohn's disease (CD). In developing countries in general and the Kingdom of Saudi Arabia (KSA) in particular, both IBD and EoE are increasingly recognized over the last few decades (El Mouzan et al., 2014; Assiri and Saeed, 2014; Hasosah et al., 2011). However, the association EoE and IBD has not been reported. The case reported here has several points of interest. First, the discovery of asymptomatic EoE on routine EGD underscores the importance of performing this procedure as part of the evaluation of patients with IBD. Second, previous reports of association were in older children whereas our patient's IBD symptoms started in infancy. Third, previous associations were with Crohn's disease and our patient had confirmed UC.

Eosinophilic esophagitis and UC are chronic inflammatory diseases. Unlike Crohn's disease, UC is characterized by superficial mucosal lesions similar to EoE but with different type of cell response being eosinophils in EoE and mixed infiltrate in UC. The predominant TH2 type response characterizes both conditions (Gupta et al., 2006). These points suggest pathogenetic similarity. However, in our patient, the absence of eosinophils in the colonic mucosal biopsies indicates separate entities not consistent with the concept of primary eosinophilic infiltration of the gastrointestinal tract (Alfadda et al., 2011).

CONCLUSION

This report is the first case of association of asymptomatic EoE and very early-onset UC. More studies in the form of genetic analysis and cytokine profiles may clarify the significance of this association.

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