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Yutma Güçlüğünün Nadir Bir Nedeni Eozinofilik Özofajit: İki Olgu Sunumu ve Literatür Derlemesi

Eosinophilic Esophagitis as a Rare Cause of Dysphagia: Two Case Presentation with Literature Review

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ÖZ

Eozinofilik özofajit (EÖ) özofagusun kronik, immün aracılı bir hastalığıdır ve genellikle özofageal disfonksiyon, retrosternal yanma, ve eozinofil baskın inflamasyon ile karakterizedir. EÖ'li birçok hasta çoğunlukla yutma güçlüğü, reflü benzeri şikayetler ve yutma güçlüğü ile prezente olur. Her ne kadar diyetsel alerjenler patofizyolojide çoğunlukla suçlansa da bilinmeyen birçok faktörün de patofizyolojiden sorumlu olduğuna inanılmaktadır. Biz bu vaka raporlarında kliniğimize yutma güçlüğü ile başvuran ve EÖ tanısı alan 2 hastayı sunmayı amaçladık. Tedavide oral flutikazon ve diyet kısıtlaması her iki hastaya da başlandı ve izlem sürecinde her iki hastada başarılı sonuçlar elde edildi.

Anahtar Kelimeler: *Eozinofilik özofajit, disfaji, steroid*

ABSTRACT

Eosinophilic esophagitis (EO) is a chronic, immune mediated disease of esophagus characterized with esophageal dysfunction, retrosternal heartburn, and eosinophil-predominant inflammation. Most patients with EO clinically presents with dysphagia, reflux-like symptoms and swallowing difficulties. Although food allergens are mostly accused in the pathophysiology of EO, there is still unknown additional factors that are belived to be exist in the pathophysiology. Herein, we would like to present two patients who admitted to our clinic with the complaint of dysphagia and was diagnosed as EO. Oral fluticazone treatment was started with dietary restriction to both patients. A favorable outcome was achieved during follow-up.

Key words: *Eosinophilic esophagitis, dysphagia, steroid*

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INTRODUCTION

Eosinophilic esophagitis (EE) is a chronic immune system mediated disease, which frequently leads to swallowing difficulty and esophageal obstruction. The basic pathophysiological mechanism is esophageal dysfunction due to eosinophilic infiltration into the esophageal mucosa (1). Although EE was not clearly defined until the 1970s, diseases related to increased eosinophil count in various segments of the gastrointestinal system have been known for a long time. EE was first defined by Landres et al. in a patient with achalasia resistant to treatment in 1978 (2). Since then, it has been defined in children and adults with increasing incidence.

There is no study on the incidence of eosinophilic esophagitis in the normal population in our country; however, the incidence of EE among patients searching for medical advice due to esophageal symptoms has been reported to be 2.6% (3). Although an increased rate of diagnosis has been made with increased knowledge on the disease, findings suggestive of EE can easily be overseen in case endoscopy is performed by physicians not experienced on this disease. Therefore, in cases with complaints suggestive of esophageal pathologies such as swallowing difficulty or feeling of retrosternal obstruction, it is more important to evaluate the esophagus carefully. In this case report, we aimed to present two cases with long-term complaints of swallowing difficulty and retrosternal obstruction, who were endoscopically and pathologically diagnosed as having EE, and who could not be diagnosed previously despite previous endoscopies.

CASE REPORT

Case 1: A 51-year-old male patient presented to our gastroenterology clinics due to swallowing difficulty and feeling of retrosternal obstruction persisting for 4-5 years. He experienced his complaints every day and did not benefit from the proton pump inhibitor and/or antacid drugs he received at various times. He had undergone endoscopy once at an external medical center within the latest 5 years and no significant pathology had been reported. He described no weight loss, vomiting or hematemesis. Physical examination was normal. No abnormality was determined in the complete blood count or the biochemistry parameters. His endoscopy revealed longitudinal linear linings **(Figure 1)** and white papules of milimetric size in one place or another within the esophagus.

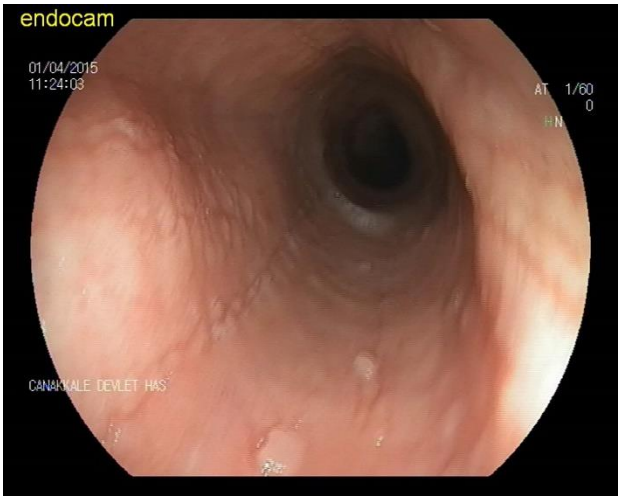

Figure 1. Linear furrows in esophagus

Diffuse eosinophilic infiltration (>30 eosinophils in each area of large magnification) was determined in the mucosa in biopsies obtained from all four quadrants of the eseophagus. The patient was diagnosed as EE and fluticasone 125 mcg inhaler 3x3 puffs (swallowing was recommended) was begun. The patient was informed regarding the elimination diet as well (that wheat, egg, soy, cow milk protein, sea food and peanut should be avoided). His complaints disappeared in the 6th month of therapy.

Case 2: A 68-year-old female patient was directed to our clinics from an external clinic with the complaint of long-term swallowing difficulty. She had not undergone endoscopy previously. Physical examination was normal except for a mild tenderness in the upper right quadrant. Complete blood count and routine biochemical parameters were normal. Her endoscopy revealed linear linings from the proximal esophagus to the distal part, and papules of milimetric size in one place or another **(Figure 2).** Four biopsies were obtained from different quadrants of the eseophagus. Pathological examination revealed diffuse eosinophilic infiltration (>30 eosinophils in each area of large magnification) in the mucosal biopsy samples.

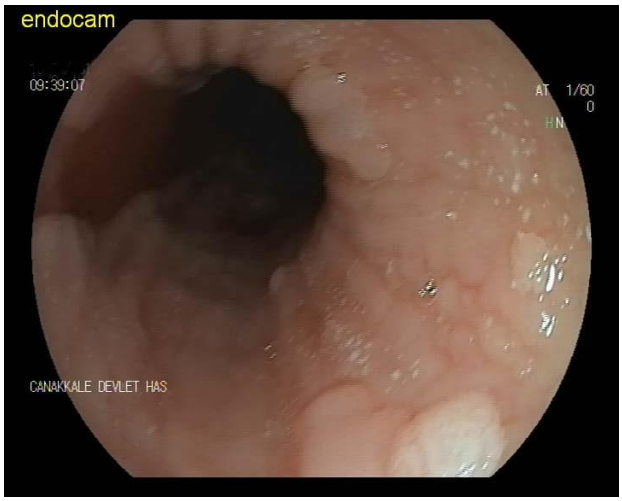

Figure 2. Linear furrows and papules

Fluticasone 125 mcg inhaler 3x3 puffs (swallowing was recommended) was begun for the patient and she was summoned for a control visit 3 months later.

DISCUSSION

In this case report, we aimed to highlight the importance of considering rare diseases such as EE in patients partially responding or not responding to the treatments they receive due to complaints such as swallowing difficulty, feeling of retrosternal obstruction or reflux. Furthermore, we emphasized the importance of endoscopic examination in the diagnosis, and elimination of alternative diagnoses.

EE is a chronic disease of the esophageal mucosa characterized by isolated eosinophilic infiltration of the mucosa (4). It is generally irresponsive to acid suppression treatments, and well responsive to steroid treatment and elimination of nutrition including dietary antigens (5). The reported incidence differs among studies; however, it was reported in a Swedish study of more than 16 years that the incidence of the disease was 1,4/100.000, and the prevalence was 2/100.000 (6). Although the number of studies on this disease is on the increase with the increase in the knowledge in our country, most of these studies are case reports (7,8). The first study investigating the incidence of EE in adult patients was conducted by Altun et al. in 2013 in our country (3). Multiple biopsies were obtained from different segments of the esophagus in 311 patients for a 2-year period due to esophageal symptoms, and histopathological examination was performed. Pathological findings compatible with EE were observed in 8 of these patients (2.6%).

There are some endoscopic findings that are not specific to EE but are common in the disease. These findings include esophageal rings, linings, papules and strictures. In both of our cases, esophageal linear linings and papules of milimetric size were present in the endoscopic images. These images were suggestive of EE and multiple biopsies were obtained for the diagnosis. In the study of Altun et al., the most common endoscopic findings among patients with EE were esophageal rings, white exudate and linear linings. A normal endoscopic image was also observed in some of the cases (3). Diagnostic histopathological findings of EE include >20 eosinophils in each area of large magnification in mucosal biopsy samples, presence of eosinophilic micro-abscess, stratification of eosinophils along the surface of the esophagus wall, and basal regional hyperplasia (1). The findings of our cases were compatible with those observed in the literature histopathologically.

Medical treatment of EE includes elimination of allergic nutrient from the diet both in children and in the adults. Therefore, 6 different types of food that have the highest risk of exacerbating symptoms are recommended to be avoided in EE. These are: wheat, egg, soy, cow milk protein, seafood and peanut. An acceptable response is obtained with the elimination of these foods from diet in children; however, a lower rate of response is obtained in adults (9,10). Topical steroids are primarily preferred in the treatment of adults; however, the possibility of recurrence should always be considered (11). In patients with insufficient response to topical therapy, systemic corticosteroids may be used. Agents such as montelucast and mopelizumab may be used clinically as well. In patients irresponsive to medical treatment and with esophageal stricture, plug or balloon dilatations may be placed endoscopically (9). Since there was no endoscopically detected stenosis in any of our cases, we primarily performed a limited diet and topical steroid use. No systemic therapy was planned due to the significant response obtained from the treatment.

In conclusion, EE should be considered in patients with esophageal symptoms who are irresponsive to and/or partially responsive to standard antacid and proton pump inhibitor agents. Elimination of the disease pathologically with a minimum of 4 biopsies obtained from different sites of the esophagus is important, even in case of normal mucosal appearance on endoscopic examination.

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