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Original Article

An Uncommon Cause of Dysphagia

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ABSTRACT

Dysphagia is a frequent indication for a referral to a gastroenterologist. The most common etiologies of dysphagia include gastroesophageal reflux disease, eosinophilic esophagitis (EoE), or structural disorders of the esophagus. We present a case of refractory dysphagia in a patient with a history of Sjogren's disease and autoimmune hepatitis. Upper endoscopy showed crepe paper appearance of the esophagus raising suspicion for EoE; however, biopsies revealed lymphocytic esophagitis (LE). As LE is a rare entity, optimal treatment remains unknown. Nonetheless, applying the common methods used to treat EoE including proton pump inhibitors, topical steroids, and esophageal dilations improved the patient's symptoms.

Key words: Esophagus, Lymphocytic esophagitis, Eosinophilic esophagitis, Upper endoscopy, Esophageal dilation..

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INTRODUCTION

Dysphagia is a common symptom that is evaluated in ten million Americans yearly.[1] A variety of etiologies can result in dysphagia including esophageal lumen narrowing by inflammation, stricture, web, or tumor as well as motility disturbances.[2] While the most common causes of inflammation leading to dysphagia are gastroesophageal reflux disease (GERD) and eosinophilic esophagitis (EoE), other etiologies exist. We present a case of an unusual etiology of dysphagia.

CASE REPORT

A 37-year-old woman with a history of Sjogren's disease and autoimmune hepatitis presented for evaluation of dysphagia to solid food for several years. Prior upper endoscopy showed LA grade A esophagitis for which she was treated with proton pump inhibitor (PPI). She remained on this therapy for two years; however, her dysphagia persisted. She ultimately sought care in our clinic for PPI-refractory dysphagia.

An upper endoscopy was repeated, which showed circumferential esophagitis in the distal one third of the esophagus as well as an esophageal narrowing at 25 cm from the incisors (Figure 1). The esophagus was friable and bled with contact of the endoscope (Figure 2). Because of crepe paper appearance of esophagus, there was a high suspicion for eosinophilic esophagitis (EoE). However, the biopsies revealed ~146 intraepithelial lymphocytes within squamous mucosa in 1 high power field (146 IEL/HPF), and no eosinophils were present. These findings were consistent with lymphocytic esophagitis (LE). The patient was prescribed topical fluticasone for LE in addition to twice daily PPI. The steroid course was briefly interrupted by an episode of candida esophagitis which was treated with oral fluconazole. The patient reported overall improvement in her symptoms with these therapies.

Two years later, the patient presented to the emergency room with a food impaction. An upper endoscopy was performed to remove the bolus and it revealed a stricture at 25cm from the incisors estimated to be approximately 8mm in diameter. Subsequently two upper endoscopies were performed for dilation of the known stricture. The first dilation was up to 11mm and the biopsies taken showed a peak count of 70 IEL/HPF. The second dilation was performed 3 months later, and it was up to 12mm (Figure 3). Appearance of the mucosa was bland endoscopically without overt inflammation. The friability and crepe paper appearance of the mucosa had resolved (Figure 3). Biopsies from most recent endoscopy demonstrated only scattered lymphocytes and a few (8/HPF) eosinophils. After most recent dilation, patient reported complete resolution of dysphagia.

DISCUSSION

LE is a rare entity that is detected in only 1/1000 esophageal biopsies but is becoming increasingly recognized as a cause of dysphagia.[3] LE is characterized by an increase in the number of intraepithelial lymphocytes,[4] though the exact count of lymphocytes needed for the diagnosis remains unclear.[5] Patients with LE typically present with dysphagia, chest/abdominal pain, and acid reflux.[6] Moreover, LE occurs more frequently in women with a history of immune-mediated disorders than in men.[7] Effective treatment for LE remains unknown,[8] though treatments that have been implemented with some success include those used for EoE such as PPI, steroids, and serial esophageal dilations.[9]

We initially chose to treat this patient with topical fluticasone 440mcg twice daily. This therapy was complicated by an episode of candida esophagitis, yet

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the patient was ultimately able to resume treatment while continuing on a PPI. The patient also underwent multiple esophageal balloon dilations for persistent stricture despite medical therapy. Subsequent biopsies showed a continuous reduction in the count of intraepithelial lymphocytes and the patient reported resolution of symptoms after her most recent dilation. While topical fluticasone did not result in complete histologic healing of the esophageal mucosa, there was a marked decreased in lymphocytes over time with this therapy.

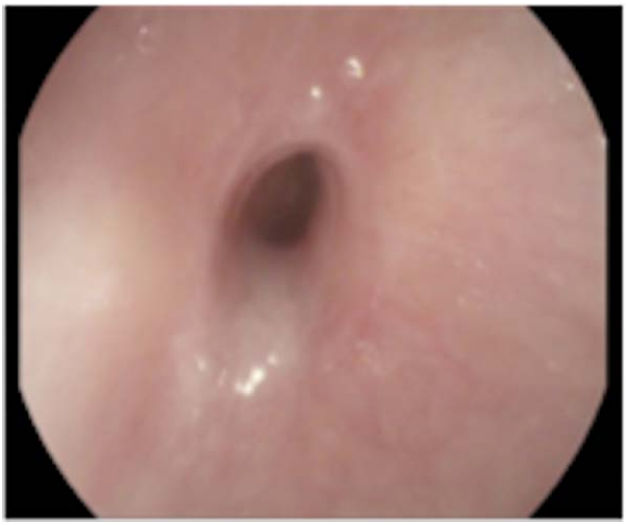

Figure 1. Endoscopic appearance at diagnosis showing esophageal narrowing at 25cm.

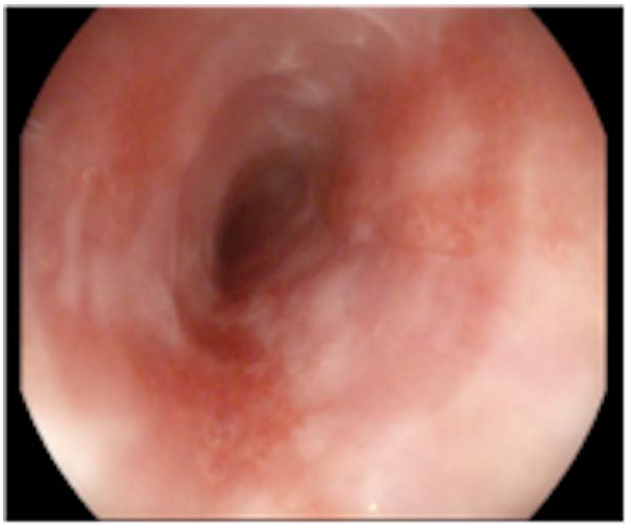

Figure 2. Esophagitis characterized by edema and friability at time of diagnosis.

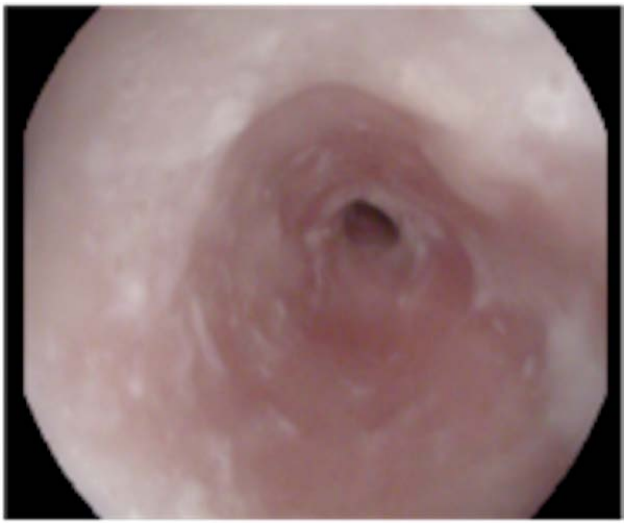

Figure 3. Esophagus with bland appearance post-treatment with topical steroids.

In conclusion, LE should be considered as a potential cause of dysphagia and esophageal inflammation, especially in patients with underlying autoimmune disease. More research is needed to determine optimal treatment regimen for LE, but the combination of PPI and topical steroids for mucosal inflammation in addition to esophageal dilations for persistent stricture was a successful approach for this patient with LE.

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