# Lymphocytic oesophagitis: an emerging cause of dysphagia

## Introduction

A 58-year-old woman with a several year history of dysphagia presented with recurrent oesophageal stricturing despite multiple gastroscopic dilations. Investigative endoscopy revealed a 'ridged' oesophagus with prominent rings in the middle third, raising a suspicion of eosinophilic oesophagitis. Interestingly biopsies in fact demonstrated a marked lymphocytic inflammatory cell infiltrate and an absence of eosinophils, so a diagnosis of lymphocytic oesophagitis was made. Treatment with steroids resulted in symptomatic relief mirroring both macroscopic and microscopic improvement of the oesophagus.

This report describes a newly emerging condition, and is unique because of the context of the patient's family history. Her mother had been diagnosed with lymphocytic oesophagitis 1 year previously, and while there are currently no data regarding the role of genetics in its pathophysiology these two related cases raise the possibility of a genetic component.

## Discussion

Lymphocytic oesophagitis is a newly emerging condition first reported by Rubio et al in 2006, yet remains relatively poorly understood. It presents similarly to eosinophilic oesophagitis with dysphagia, oesophageal rings and stricturing, but histologically the condition is characterized by marked oesophageal lymphocytosis and an absence of eosinophils (Rubio et al, 2006). The predominantly peripapillary distribution of the lymphocytes also notably differs from other established causes of oesophagitis (e.g. reflux, radiation, *Candida albicans*), in which intraepithelial lymphocytes are mostly identified in an interpapillary distribution.

Fewer than fifty cases of lymphocytic oesophagitis have been reported in the literature to date and there remains little research regarding patient demographics or associated conditions (Haque and Genta, 2012; Pasricha et al, 2016; Pleet et al, 2017). A single study investigating the clinical presentation and natural history of 81 patients diagnosed over a 10-year period found the condition to be more common in women and overwhelmingly in the Caucasian population (Cohen et al, 2012). It is typically diagnosed between the ages of 40 and 59 years, but occurrences have been noted in paediatric populations, including cases of children with Crohn's disease (Ebach et al, 2011; Sutton et al, 2014).

Figure 1. Ridged oesophagus with prominent rings, middle third.

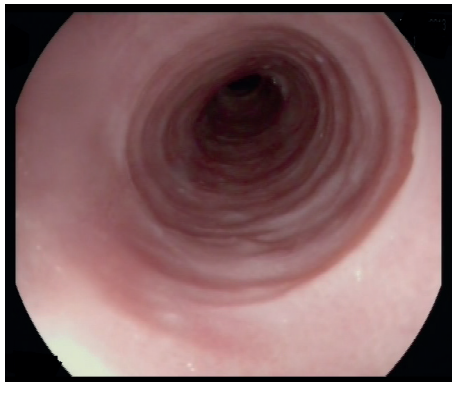
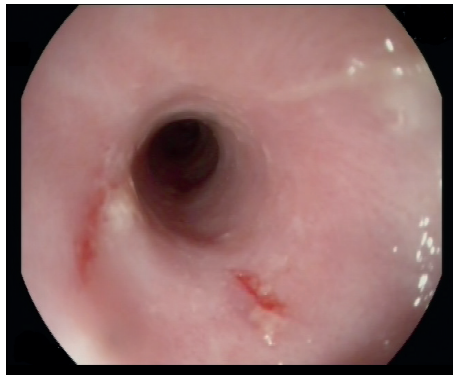


Figure 2. Fragile oesophageal mucosa.



## CASE REPORT

A 58-year-old woman was referred to the gastroenterology clinic with a several year history of high oesophageal dysphagia. She described episodes of food 'sticking', with significant psychological impact, despite five attempted dilations for her known oesophageal webs under the ear, nose and throat department. Her past medical history included vitamin $B_{12}$ deficiency, eczema, a hysterectomy and a Mallory–Weiss tear. Of interest the patient's 82-year-old mother had been diagnosed with lymphocytic oesophagitis within the last year following an 8-year history of dysphagia. At the time of presentation neither this condition nor its 'sister condition' eosinophilic oesophagitis had been investigated for in the patient.

Endoscopy revealed a distinctive 'ridged' oesophagus, with prominent rings demonstrated in its middle third (*Figure 1*). The mucosa was fragile and stripped easily to minimal trauma and in response to biopsies (*Figures 2* and *3*). A diagnosis of eosinophilic oesophagitis was suspected macroscopically until mid-oesophageal biopsies demonstrated squamous cell mucosa with a striking lymphocytic inflammatory cell infiltrate. The infiltrate was centred around the papillae and associated with epithelial spongiosis. These features are typical in lymphocytic oesophagitis, and given the lack of an eosinophilic component a diagnosis of eosinophilic oesophagitis could be excluded.

Following the diagnosis of lymphocytic oesophagitis, the patient was commenced on a course of inhaled corticosteroids (fluticasone propionate; Flixotide), with subsequent symptomatic improvement. Repeat endoscopy 6 months later revealed no remaining stricturing or ridging. Biopsies demonstrated a comparative reduction in lymphocytic infiltration and almost complete resolution of any acute inflammatory component. Oral corticosteroids were prescribed for full symptomatic control.

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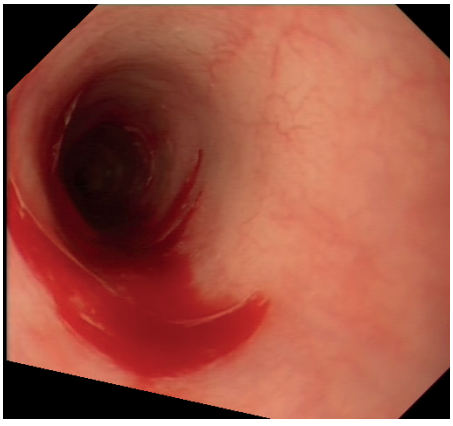
Figure 3. Oesophageal mucosa easily stripped following minimal trauma and biopsies.



The 'sister condition' eosinophilic oesophagitis is commonly linked to allergy or atopy, with one case series demonstrating that 36% of patients had asthma, 54% had peripheral eosinophilia and 10% had a first-degree relative with dysphagia (Pasha et al, 2007). However, no significant association between lymphocytic oesophagitis and atopy has currently been established and no familial links have been reported (Purdy et al, 2008).

This case reports a new condition that may be easily missed without a high index of suspicion. Current treatment is not well researched, but it is likely to respond to steroids and there are suggestions that immunosuppression may be beneficial in this evolving field (Akbar et al, 2016). This unique finding in two first degree relatives may open the discussion for a possible genetic component to the condition. **BJHM**

## LEARNING POINTS

- Dysphagia can present via many pathways, from general internal medicine to ear, nose and throat and general practice, and it is important to biopsy patients with this presentation.
- Oesophageal strictures may be malignant, peptic or non-peptic inflammatory.
- Lymphocytic oesophagitis presents similarly to eosinophilic oesophagitis but may not be recognized clinically or histologically as an entity.
- Both lymphocytic oesophagitis and eosinophilic oesophagitis can be treated with steroids.
- This unique finding in two first degree relatives opens the discussion for a possible genetic component to the condition.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