

## **REPORTE DE CASO**

# Ringed esophagus: all that glitters is not gold Anillos esofágicos: no todo lo que brilla es oro

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JFB: Development of the manuscript concept. Literature review. Draft writing and final version. ABC: Development of the manuscript concept. Literature review. Writing of the final version. Format and style evaluation. MT: Development of the manuscript concept. Literature review. Writing of the final version. Format and style evaluation.

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#### ABSTRACT

We report the case of an elderly patient with progressive dysphagia to solids and later to liquids, and weight loss. The patient underwent an upper endoscopy, which showed multiple stenoses and trachealization. Biopsies were taken and a diagnosis of lymphocytic esophagitis was made. Pneumatic dilation to 12 mm was performed, and the patient recovered with no recurring symptoms. Dysphagia in the elderly should not be overlooked, and physicians should consider lymphocytic esophagitis as a differential diagnosis.

**Keywords:** Esophagitis; Dysphagia; Intraepithelial Lymphocytes; Endoscopy (source: MeSH NLM).

#### RESUMEN

Reportamos el caso de un paciente adulto mayor con disfagia a sólidos que progresó a líquidos, con pérdida de peso asociada. Se hizo una endoscopía alta, que mostró múltiples estenosis y traquealización del esófago. La evaluación anatomo-patológica contirbuyó a un diagnóstico de esofagitis linfocítica. Se hizo una dilatación neumática hasta 12 mm, y el paciente se recuperó sin recurrencia de síntomas. La disfagia en el adulto mayor no debe pasarse por alto, y los médicos deben considerar la esofagitis linfocítica como un diagnóstico diferencial.

**Palabras clave:** Esofagitis; Disfagia; Linfocitos Intraepiteliales; Endoscopía (fuente: DeCS Bireme).

## INTRODUCTION

Since its recognition in 2006, lymphocytic esophagitis has emerged as a cause of chronic dysphagia in the elderly. It is important that treating physicians consider it as a differential diagnosis in the evaluation of dysphagia and be familiar with the clinical presentations, gross endoscopic findings, the need for biopsy for histopathological evaluation, and management of common complications. Also, they should consider the similarities and differences with eosinophilic esophagitis, which have implications in evaluating and managing these conditions. We present the case of a patient with lymphocytic esophagitis.

## **CASE REPORT**

An 81-year-old male patient with a history of hypertension and benign prostatic hyperplasia was seen in the clinic complaining of progressive and worsening dysphagia for the last 18 months. The dysphagia was first for solid foods and over the previous 6 months, it included liquids too. The patient complained of a 10 kg weight loss for the previous 4 months. History was negative for heartburn, gastroesophageal reflux, chest pain, rash, arthralgia, fever, and respiratory or systemic symptoms. His only medications were losartan 50 mg and tamsulosin 0.4 mg, both taken once daily. There is no history of allergies. The patient denied the use of alcohol or tobacco. There is no family history of neoplasia or autoimmune conditions.

At the first evaluation, the patient was hemodynamically stable and without any relevant findings in the physical exam. Laboratory results showed no abnormalities. Hemoglobin was 13.1 g/dL.

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Jeffrey Freidenson Universidad Peruana Cayetano Heredia Av. Honorio Delgado 430, San Martín de Porres. Lima, Perú. E-mail: jeffrey.freidenson.b@upch.pe Given his age, symptoms, and weight loss, an esophagogastroduodenoscopy (EGD) was performed (Figure 1 A, B, and C). The EGD revealed multiple stenoses in the lower half of the esophagus, partial trachealization with ring formation, and edema. Proximal stenosis allowed endoscope passage with moderate resistance and minimal bleeding. A distal stenosis 36 cm from the dental arch, less than 10 mm wide, prevented endoscope passage.

During the EGD, two biopsies from the middle and distal esophagus were taken. Histopathological analysis revealed squamous epithelium with multiple intraepithelial lymphocytes in both samples (Figure 1 D and E). Figure 1 E shows a magnified view of the biopsy, with intraepithelial lymphocytes indicated by the black arrows, and dyskeratotic cells of the esophageal epithelium inside the blue circles.

The stenosis was pneumatically dilated until passage to the stomach was achieved at 12 mm. The stomach and duodenum showed no significant alterations. The patient tolerated the procedure well, including the pneumatic dilation. A month following the procedure, the patient was asymptomatic.

## DISCUSSION

Dysphagia can be considered a geriatric syndrome, as it affects 10-33% of older adults <sup>(1)</sup>. Aging is associated with structural and physiological changes impairing swallowing,

including reduced contraction of esophageal smooth muscle, nonperistaltic contractions, and delayed esophageal emptying, among other processes <sup>(1)</sup>. However, dysphagia should not be considered normal, and its presence in the context of other symptoms warrants investigations by the treating physician.

Complaints of retrosternal sensation of bolus, chest pain, and dyspepsia after swallowing point towards esophageal dysphagia. Esophageal dysphagia has a variety of etiologies. Intrinsic mechanical causes include esophageal rings and webs (Shatzki, Plummer-Vinson), strictures from multiple causes (GERD, scleroderma, caustic injury, post-surgical), neoplasm (squamous cell carcinoma, adenocarcinoma), diverticula, benign masses, among others. Extrinsic mechanical causes include mediastinal mass (lymph nodes, thyromegaly, lung cancer) and vascular compression (enlarged left atrium, aberrant right subclavian artery, right-sided aorta). Neuromuscular causes include achalasia, scleroderma, hypercontractile peristalsis (nutcracker esophagus), hypertensive lower esophageal disorder, Chagas disease, paraneoplastic syndromes; etc. Inflammatory causes include eosinophilic esophagitis (EoE), radiation esophagitis, pill-induced esophagitis, and infectious etiologies (cytomegalovirus, candidiasis, herpes simplex, or HIV-associated esophagitis) <sup>(2)</sup>.

In our case, the patient had progressive dysphagia to solids and later to liquids, suggesting a worsening mechanical obstruction, and a 10-kg weight loss over 4



Figure 1. Pictures of esophagogastroduodenoscopy and histopathology analysis of esophageal biopsy samples. A) middle esophageal stenosis with edema. B) severe stenosis in the distal esophagus before pneumatic dilation. C) distal esophagus after pneumatic dilation. Of note, a mild esophageal laceration resulted from the procedure. (Courtesy of Dra Mariam Alsharif, GastroHealth EEUU).

months. This was particularly concerning for a possible neoplasm causing mechanical obstruction, and therefore an EGD was performed. The gross evaluation showed ring formation with multiple stenoses. Esophageal rings are mostly asymptomatic but can sometimes cause intermittent dysphagia to solids that slowly progress to liquids after years. Symptoms are present when the luminal diameter is less than 12 mm <sup>(3)</sup>. Esophageal rings are associated with hiatal hernia, chronic irritation caused by GERD, and eosinophilic esophagitis <sup>(3)</sup>.

Eosinophilic esophagitis is an immune-mediated condition that affects males and young people more often but can also appear in the elderly <sup>(4)</sup>. Dysphagia is the main symptom, and can also cause food impaction, chest pain, refractory heartburn, and upper abdominal pain <sup>(5)</sup>. Endoscopic findings include furrows (vertical lines in the mucosa), concentric rings with esophageal narrowing (trachealization), exudates, edema and strictures <sup>(5)</sup>. The International consensus diagnostic criteria for eosinophilic esophagitis established specific criteria to diagnose EoE. This consists of symptoms of esophageal dysfunction and at least 15 eosinophils per high-powered field (or ~60 eosinophils per mm<sup>2</sup>) on esophageal biopsy, and after evaluation of other disorders that could cause esophageal eosinophilia (6). In this case, the patient had recalcitrant dysphagia, which represents esophageal dysfunction, and the biopsy showed multiple intraepithelial lymphocytes and no eosinophils, ruling out EoE, and confirming lymphocytic esophagitis.

Lymphocytic esophagitis is a chronic inflammatory disorder of the esophagus, characterized by the presence of peripapillary intraepithelial lymphocytes in the absence or minimal presence of intraepithelial granulocytes (7) It is more prevalent in older women, and is associated with GERD, motility disorders (i.e achalasia), tobacco use, allergies, hypothyroidism and the use of certain medications like aspirin or statins <sup>(8)</sup>. Patients usually present with dysphagia, heartburn, chest pain, and upper abdominal pain, similar to EoE. Endoscopic findings are varied, ranging from a normalappearing esophagus to strictures, rings, furrows, erosive esophagitis, edema, and white plaques <sup>(9)</sup>. Even though the diagnosis is made by histopathological evaluation, in contrast to EoE, there are no standardized criteria and no agreement on the number of intraepithelial lymphocytes required for diagnosis. However, the criteria most accepted involves a pattern of 20 intraepithelial lymphocytes per highpower field, few or absent granulocytes, and spongiosis <sup>(10)</sup>. A study by Cohen et al. identified strictures in 8.3% of patients with lymphocytic esophagitis (11). In our case, the patient presented with many gross endoscopic findings including rings, edema, and a stricture with a luminal diameter of less than 10 mm (Figure 1). Therefore, endoscopic dilation was performed, and the patient recovered fully. Based on reported cases, dilation can be repeated if needed <sup>(10)</sup>. Of note, there is a lack of evidence-based clinical guidelines to treat lymphocytic esophagitis, and treatment alternatives usually mimic those for EoE, with high-dose proton-pump inhibitors (PPIs) being the first alternative. Currently, it is unclear if the reported improvement with PPIs is due to an improvement of lymphocytic esophagitis, or an improvement of concomitant GERD <sup>(12)</sup>.

In conclusion, lymphocytic esophagitis as a cause of dysphagia must be considered when evaluating patients for dysphagia, particularly in the elderly. It is necessary to take biopsies when doing an EGD to look for this condition. Patients can present with esophageal strictures and pneumatic dilation may be required.

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