

## Esophageal Web Complicating an Isolated Esophageal Lichen Planus

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**Lichen planus (LP) is a chronic, idiopathic disorder affecting the mucosal surfaces, skin and nails. Esophageal involvement in this disease is rare and only few cases were found in literature, thus making its diagnosis challenging as it can be easily misdiagnosed as gastroesophageal reflux disease. Currently, little is known about its pathogenesis and management.**

**We report a case of a previously healthy 32-year-old female who presented with the complaint of dysphagia, which was later diagnosed endoscopically as an esophageal web. Biopsy of the lesion revealed a histological diagnosis of an esophageal lichen planus (ELP). This was treated with multiple Esophagogastro Duodenoscopy (OGD) dilatation sessions and local steroids. We also reviewed similar reported cases in the literature, stressing on the importance of the successful management of such a disease and its complications.**

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Lichen Planus (LP) is a mucocutaneous disease of the stratified squamous epithelium, which affects the mucous membranes, skin, nails, and scalp. LP is estimated to affect 0.5% to 2.0% of the general population. It is mostly reported in middle-aged patients (between 30-60 years of age) and is seen more evidently in females<sup>1</sup>.

It is one of the chronic disorders where oral involvement may precede the appearance of other lesions at other locations. The etiology is not well known but has been linked to the autoimmune system. The chronic disease is apparently mediated by an antigen-specific mechanism, and non-specific mechanisms like mast cells degranulation<sup>1</sup>.

Some studies suggested that esophageal lichen planus (ELP) is more common than previously known and that many patients have had the disease confined to the esophagus before developing lesions elsewhere<sup>2</sup>. Given that the symptoms of esophageal lichen planus do not present in a predictable way, ELP is probably more under-diagnosed than rare<sup>3</sup>.

Barium esophagram may reveal a small caliber esophagus or, less commonly, segmental esophageal webs or strictures<sup>2</sup>. Esophageal shadows with a configuration resembling rings and webs are often described by radiologists in patients complaining of dysphagia. Those located in the upper esophagus of women and associated with iron deficiency anemia are a component of the Plummer-Vinson or Paterson-Kelly syndrome. Those located in the lower esophagus are commonly referred to as lower esophageal rings<sup>3</sup>. To date, a few cases of squamous cell carcinoma in association to ELP have been reported, indicating the importance of timely diagnosis and earlier management<sup>4</sup>.

The treatment of esophageal webs includes endoscopic dilatation, steroids and surgical myomectomy<sup>3</sup>.

The relationship between ELP and esophageal webs is not clear and rarely reported in the literature.

The aim of this presentation is to report a case of esophageal web complicating an ELP.

### THE CASE

A thirty-two-year-old, previously healthy Yemeni female presented to the gastroenterology clinic in January 2015 with complaints of dysphagia and a sensation of a foreign body in her throat. She was referred from the ENT clinic after exclusion of local cause. On clinical examination, a granular pharynx with lateral pharyngeal bands was noted.

One month later, she was still complaining of difficulty in swallowing hard food and weight loss of 12 kilograms in the last six months. Hb level, iron studies, and thyroid function tests were normal; therefore, Plummer Vinson syndrome was excluded. Her neck, lymph nodes, and genital examination were normal.

Barium swallow showed moderate reflux disease and her weight was 41.45 kilograms. She was stable symptom-wise, but the weight loss was still a concern. She was found to be positive for *H-Pylori*; however, triple therapy was not given as she was unable to swallow the tablets. OGD revealed a very narrow opening, with a localized, thick and circumferential post-cricoid web and was diagnosed accordingly with an

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oesophageal web, see figure 1. The patient was treated with mechanical dilatation during the same session.

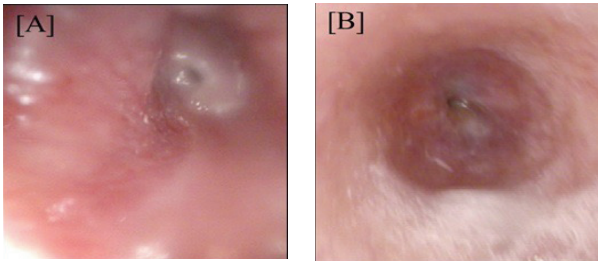


Figure 1 (A)

Figure 1 (B)

**Figure 1: (A) OGD Pre Treatment Showing Esophageal Web (B) OGD Post-Treatment Esophageal Web Resolution**

Post-dilatation, the patient had no dysphagia, gained 1 kilogram and was started on triple therapy for *H. pylori* gastritis.

However, in January 2016, her dysphagia recurred and weighed 39.5 kilograms. A second OGD revealed the recurrence of the post-cricoid esophageal web. Dilatation with 15 mm balloon was performed. In April 2016, she underwent a third OGD and a recurrence of the web was found; however, it was easily ruptured by the scope and topical steroid was used locally. A biopsy was taken for histopathology. She was prescribed Sucralfate and Avamys (one puff, twice a day).

The histopathology report described the findings of parakeratotic esophageal mucosa showing moderately severe lichenoid mononuclear chronic inflammatory cell infiltrate with lymphocytic aggregates in the subepithelial layer. Prominent intraepithelial lymphocytic infiltrate with few scattered Civatte bodies were found. Those appearances are highly suggestive of ELP and reflux esophagitis was excluded, see figure 2.

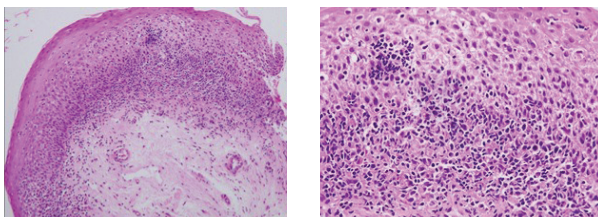


Figure 2 (A)

Figure 2 (B)

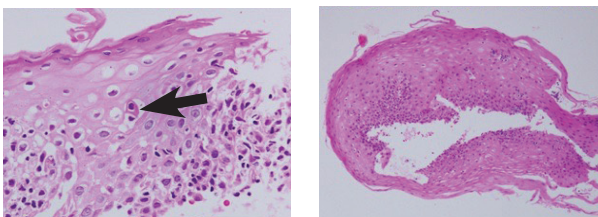


Figure 2 (C)

Figure 2 (D)

**Figure 2:**

- (A) Esophageal Biopsy Covered by Stratified Squamous Epithelium with Dense Sub-Epithelial Chronic Inflammation
- (B) High Power View of A
- (C) Apoptotic Bodies (Arrow)
- (D) Post Treatment: Esophageal Biopsy Lined by Stratified Squamous Epithelium with Resolving Inflammation

The patient reported improvement with the use of Fluticasone nasal spray and was advised to continue the same management until her fourth OGD session. The fourth OGD revealed a thin, easily ruptured esophageal web with mild candidiasis, which was treated with Fluconazole and Fluticasone. In May 2017, the fifth OGD revealed complete resolution of the web and the patient reported to be symptom-free. The patient gained 5 kilograms. Currently, the patient is doing very well and is still being followed closely by the gastroenterology team, with no evidence of recurrence of her dysphagia or the web.

**DISCUSSION**

Lichen planus is a common disorder of the squamous epithelium. The exact cause is unknown; however, viral infections, genetic and immunologic causes may have a role in the pathogenesis<sup>5</sup>. It seems to be mediated by cytotoxic CD8+ T cells that attack an antigen in the basal epithelium, resembling the process noticed in graft-versus-host disease. Oral lichen planus might be due to an immune response to an exogenous or endogenous antigen in the basal keratinocytes, which respond to injury by producing cytokines and stimulating the CD8+ T lymphocyte, leading to the distribution of the CD8+ cytotoxic cells near the epithelium and finally destroying it<sup>5</sup>.

The true prevalence of ELP is hard to determine due to the common misdiagnoses as reflux esophagitis and few reported cases<sup>5</sup>. One case series suggested that women account for almost 93%<sup>6</sup>. ELP is also described, but rarely in males<sup>5,7</sup>.

Almost 50% of patients with mucocutaneous lichen planus had esophageal involvement<sup>5</sup>. Symptoms can range from none to odynophagia and dysphagia, mostly affecting the proximal or mid-esophagus. Lesions ranging from elevated lacy white papules, esophageal webs, pseudo-membranes, erosions, and strictures have been reported<sup>5</sup>.

Esophageal webs and rings are described as thin mucosal membranes that project into the esophageal lumen. Microscopically, they consist of squamous mucosa, sometimes with acanthosis or hyperkeratosis, covering a thin layer of fibrous tissue. They may occur at the upper esophagus, where they may be related to GERD. Webs may form as adverse side effects due to radiotherapy causing epithelial injury or graft-versus-host disease<sup>8</sup>.

There are no clear guidelines to treat such cases. Systemic corticosteroids have been used as first-line treatment. Topical steroids have been used recently; however, results vary between patients<sup>7</sup>. Esophageal dilatation is an accepted procedure for the treatment of esophageal strictures or webs. Intralesional steroid injections, inhaled steroid and/or Tacrolimus have been used<sup>2,7</sup>. Stein et al demonstrated that a removable stent can be added to the list of possible treatment options after standard medical therapy and endoscopic dilatation had failed<sup>6</sup>. Ynson et al reviewed and reported a case series where treatment with swallowed fluticasone propionate resulted in satisfactory outcome<sup>5</sup>. Esophagectomy was performed by Bharat Rao et al in a female patient with a severe stricture of ELP who failed multiple courses of intralesional, topical and systemic glucocorticoid therapies with multiple endoscopic dilatation sessions<sup>9</sup>.

Oral lichen planus has 1-3% risk of squamous cell carcinoma, yet this is still unknown for ELP<sup>4,5</sup>.

## CONCLUSION

**The formation of esophageal web or stricture is a rare complication of ELP. ELP must be suspected in middle-aged females presenting with symptoms of odynophagia and dysphagia. A detailed history, physical examination, endoscopy and biopsies are essential for diagnosis. The physician should be aware about the relationship between ELP and esophageal webs as proper management is essential to prevent recurrence of the disease and its possible progression to malignancy.**

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