Dysphagia, rings and furrows; oh my! A rare cause of benign esophageal stenosis.



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Background

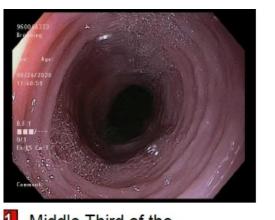
- Lichen planus (LP) is a chronic, inflammatory autoimmune disease that affects the skin, nails and mucosal membranes.
- Mucosal lesions occur in 30-70% of patients without skin lesions.
- LP is a rare cause of esophagitis and stricture/stenosis which requires esophageal dilation.

Purpose

- Discuss the presentation of esophageal lichen planus
- Review the endoscopic and histologic findings associated with esophageal LP
- Discuss the management of esophageal LP including PPI therapy and esophageal dilation

Case Description

- 52 year old female presents for second opinion of progressive dysphagia to solids for 18 months.
- She complains of feeling like she is "swallowing a golf ball" with slow transit, odynophagia and pills getting stuck. She also complains of a burning sensation with carbonated drinks, wine and spicy food. She experiences heartburn three time per week with with nocturnal symptoms.
- 20 pound weight loss over 3-4 months.
- PMH includes GERD, bronchiectasis, no significant family hx.
- She was previously on a PPI but currently unable to swallow pills
- Physical exam unremarkable and oral mucosa without ulcers or lesions and no rashes
- She was started on lansoprazole 30mg dissolved twice daily
- EGD 12 weeks later demonstrated benign appearing stenosis in the lower third of the esophagus about 8mm in diameter and less than 1 cm in length. She also had LA grade A esophagitis, atrophy, longitudinal markins, tight circumferential folds, decreased vascular pattern, whispy white areas and white specks. Esophagus was dilated to 12mm.
- Pathology demonstrated squamous cells with mixed inflammatory cells including lymphocytes and eosinophils (up to 12 per HPF), mid esophagus with squamous epithelium with mixed inflammatory cells + lymphocytes eosinophils up to 10 per HPF and proximal esophagus with lymphocytes and rare eosinophils.

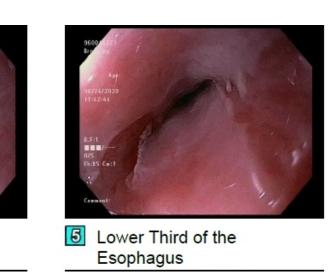




Esophagus



Esophagus



Clinical Course

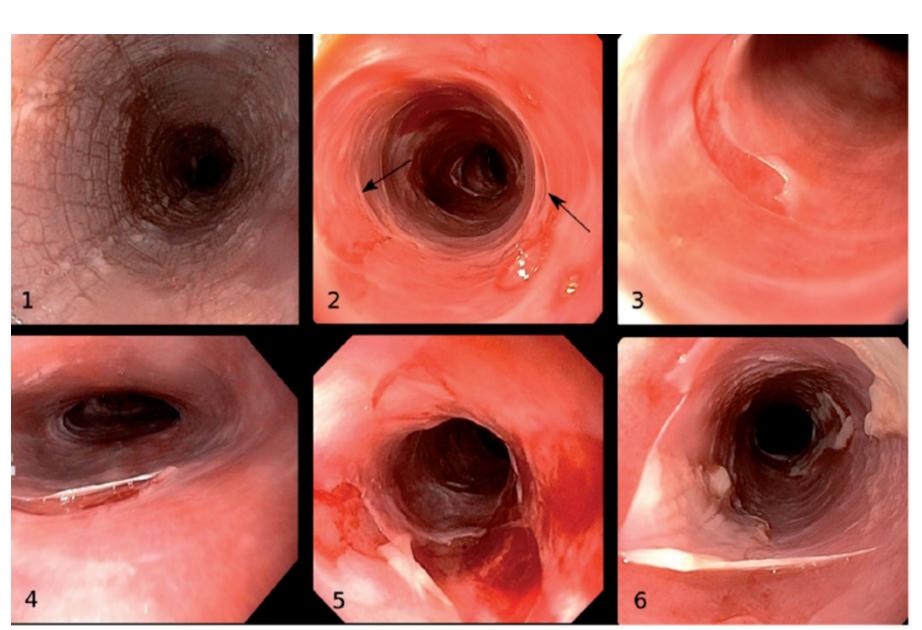
- GERD symptoms improved with PPI and dysphagia improved with dilation.
- She continued to have odynophagia with burning sensation with swallowing food
- Repeat EGD ordered off of PPI to determine if possible eosinophilic esophagitis (EoE) and decreased eosinophils due to PPI therapy.
- Repeat EGD demonstrated benign stenosis in distal esophagus and mucosal changes consistent with EoE.
- Pathology: "Basilar lymphocytosis and scattered apoptotic keratinocytes raises possibility of lichenoid esophagitis pattern which is non-specific but can be seen in association with lichen planus, medication injury/polypharmacy, viral infections and rheumatologic disorders. Patchy increased intraepithelial eosinophils which is nonspecific and seen in EoE, GERD, achalasia, drug hypersensitivity, infections"
- The patient received serial EGDs for dilation over the next several months.
- Subsequent esophageal dilation was complicated by mucosal tear requiring esophageal stenting which was eventually removed.
- She was started on swallowed budesonide to treat LP vs eosinophilic esophagitis with resolution of pills getting stuck in her throat, burning sensation and regurgitation.
- Repeat EGD after a few months of swallowed budesonide demonstrated stenosis that was dilate with Savary. Four mg of triamcinolone were injected at the site of the stricture.
- Interval between esophageal dilations has increased.
- Plans for repeat EGD with biopsies and possible repeat triamcinolone injection.

Discussion

- Esophageal involvement in lichen planus is rare. Patients often present with dysphagia or odynophagia.
- It is more common in middle-aged to older women.
- Involvement is usually in the proximal esophagus.
- Management of esophageal LP includes esophageal dilations, intralesional corticosteroids, topical corticosteroids and systemic steroids/immunosuppression.
- Refer to dermatology if there are skin or mucosal findings.

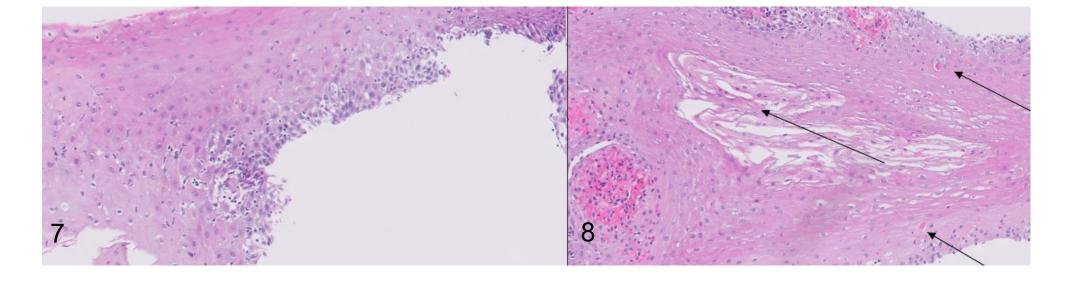
Endoscopic + Histologic Findings

- Macroscopic findings on examination include:
 - mucosal friability with denudation, sloughing and pseudomembranes
 - Hyperkeratosis with white/rough mucosal surface and plaques
 - Trachealization
 - stenosis/stricture
 - esophageal webs



- 1. Whitish, rough surface (hyperkeratosis)
- 2. Rings, trachealization 3-6. Spontaneous tearing/sloughing of mucosa

- Microscopic findings include:
 - Band-like or lichenoid lymphocytic infiltrate involving the superficial lamina propria and basal epithelium
 - Scattered eosinophilic apoptotic keratinocytes (Civatte bodies) in the basal layer
 - Predominance of CD3+ T cells in subepithelial infiltrate



7) Basal cell hyperplasia. 8) Necrotic keratinocyte (Civatte bodies).

References

Esophageal Lichen Planus: Clinical and Radiographic Findings in Eight Patients. Andreas M. Rauschecker, Marc S. Levine, Matthew J. Whitson, Rashmi Tondon, Stephen E. Rubesin, Emma E. Furth, and David C. Metz American Journal of Roentgenology 2017 208:1, 101-106

Franco et al.: Presentation, Diagnosis, and Management of Esophageal Lichen Planus: A Series of Six Cases

Vishal S. Chandan, Joseph A. Murray, Susan C. Abraham; Esophageal Lichen Planus. Arch Pathol Lab Med 1 June 2008; 132 (6): 1026–1029. doi: https://doi.org/10.5858/2008-132-1026-ELP

Steffen Kern, Adhara Lazaro, Peter Deibert, Peter Hasselblatt, Henning Schwacha, Steffen Heeg, Volker Brass, Armin Küllmer, Arthur Robert Schmidt, Annette Schmitt-Graeff & Wolfgang Ki (2019) Esophageal lichen planus: towards diagnosis of an underdiagnosed disease, Scandinavian Journal of Gastroenterology, 54:10, 1189-1198, DOI: 10.1080/00365521.2019.1674375 Kochhar, Rakesh, and Kuchhangi Suresh Poornachandra. "Intralesional steroid injection therapy in the management of resistant gastrointestinal strictures." World journal of gastrointestinal

endoscopy vol. 2,2 (2010): 61-8. doi:10.4253/wjge.v2.i2.61
"Lichen Planus, Etc.." Digital Dermatology Atlas, https://sites.uab.edu/dermatologyatlas/photo-library/lichen-planus-etc/.
Madhusudhan, K S, and Raju Sharma. "Esophageal lichen planus: a case report and review of literature." Indian journal of dermatology vol. 53,1 (2008): 26-7. doi:10.4103/0019-5154.39738
Szapáry, László et al. "Intralesional steroid is beneficial in benign refractory esophageal strictures: A meta-analysis." World journal of gastroenterology vol. 24,21 (2018): 2311-2319.