Incredibly unique case of collagenous esophagitis in a long segment Barret's esophagus: complication of chronic reflux or a new entity?

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Context

Collagenous gastroenteritides including collagenous gastritis, collagenous sprue are uncommon entities, while collagenous colitis is well known. Esophageal lamina propria fibrosis has been established in eosinophilic esophagitis as a long-term consequence of remodeling. The esophageal subepithelial collagen deposits can be regarded as collagenous esophagitis which is a new entity, which has not been described before. We herein report a case of concurrent collagenous carditis and collagenous esophagitis in a patient with Barrett's esophagus (BE).

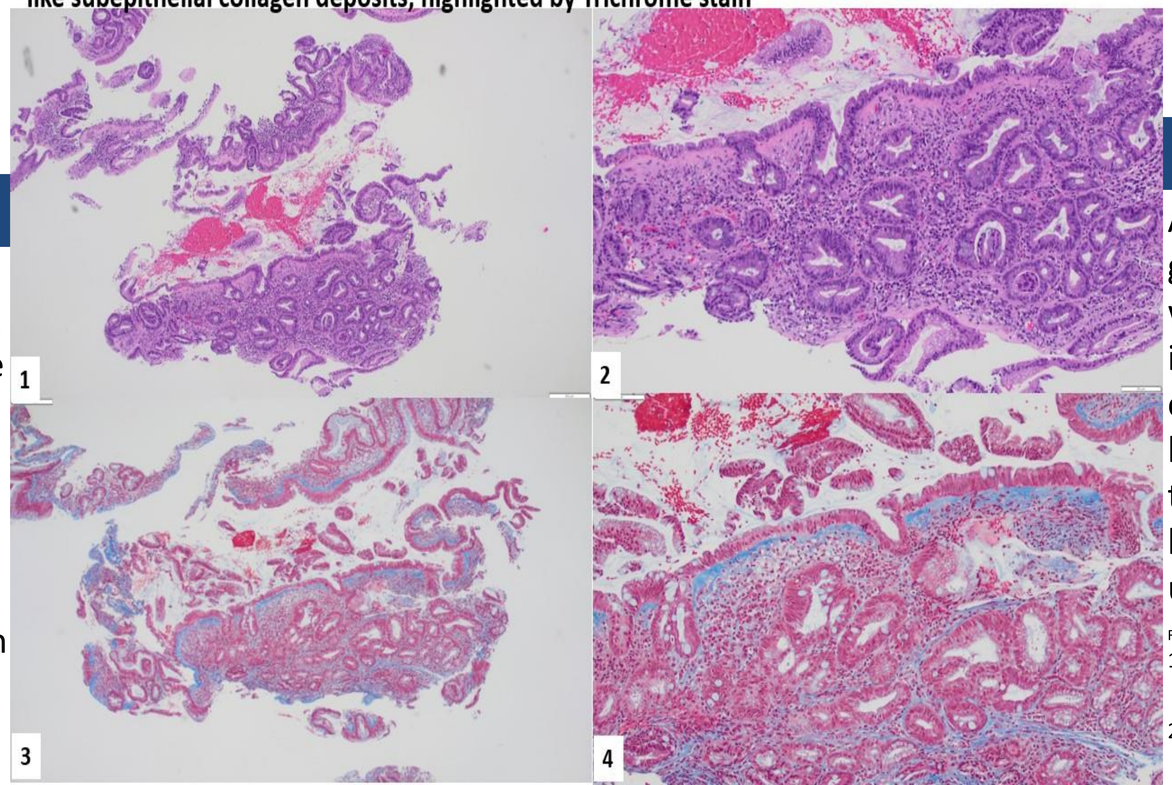
Design

36-yo male with complex medical history of cerebral palsy, seizure disorder, hypothyroidism, chronic gastroesophageal reflux disease, erosive esophagitis, who developed benign esophageal intrinsic stenosis at 23 cm from the incisors and BE in the lower third of the esophagus within ten-year period, which necessitated two endoscopic balloon dilatation procedures. Of note, he did not receive radiofrequency ablation or mucosal resection therapies at any point of time.

Results

Subsequent three endoscopies and esophageal and cardiac biopsies away from the stricture (between 25-33 cm from the incisors) demonstrated BE indefinite for dysplasia. The most recent one in addition to intestinal metaplasia had a new finding of prominent lamina propria fibrosis with diffuse band-like subepithelial collagen deposits more than 10 µm in thickness, confirmed by Trichrome stains. Congo red stains were negative. Features of eosinophilic esophagitis were absent. These findings were compatible with collagenous esophagitis and collagenous carditis diagnosis (See Figure 1).

Figure: 1) Esophageal biopsy, H&E, 20X; 2) Esophageal biopsy, H&E,100X; 3-4) Diffuse light blue band-like subepithelial collagen deposits, highlighted by Trichrome stain



Discussion

Collagenous gastritis is an extremely rare and poorly understood process with female and young adult predominance. The adult-onset collagenous gastritis is antrum dominant and is associated with collagenous colitis and autoimmune disorders, such as celiac disease, thyroid disorder, and others.

Our patient had collagenous carditis with collagenous esophagitis in the background of BE, which has never been described before.

Conclusion

Approximately 60 cases of collagenous gastritis have been reported since it was first described in 1989. An incidental discovery of subepithelial collagen deposition in our esophageal biopsy prompted an investigation into the clinical setting, review of relevant literature, and formation of this unique case report.

RREFERENCE:

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