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Glycogenic Acanthosis: An Unusual Cause of Vocal Fold Leukoplakia

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Case Report

Glycogenic Acanthosis: An Unusual Cause of Vocal Fold Leukoplakia

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Glycogenic acanthosis is a common benign lesion of the esophagus; however, reports of extra-esophageal manifestations are exceedingly rare. This case represents the first report of laryngeal glycogenic acanthosis found in a living patient, presenting as vocal fold leukoplakia. Glycogenic acanthosis may be considered among the differential diagnoses of conditions presenting as vocal fold leukoplakia.

Key Words: Glycogenic acanthosis, leukoplakia, benign vocal fold lesion.

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INTRODUCTION

Glycogenic acanthosis is a common benign lesion of the mid-to-lower esophagus. It may be seen during upper gastrointestinal endoscopy in anywhere from 3.5% to 28.3% of cases.[1,2] It tends to grossly appear as light-colored, multifocal polyps or plaques involving otherwise normal esophageal mucosa, and it is characterized histologically by hyperplastic squamous epithelium with abundant intracellular deposits of glycogen.[3] Esophageal glycogenic acanthosis was first described by Rywlin and Ortega in 1970. Little is known about its etiology or clinical significance, but there has been some evidence of an association with esophagitis and gastroesophageal reflux disease (GERD)[1,2] and, more recently, with metabolic syndrome and insulin resistance.

Despite its frequent occurrence in the esophagus, reports of extra-esophageal manifestations are exceedingly rare.[4] To date, there has only been one case of laryngeal glycogenic acanthosis described in the literature, which was incidentally found in an autopsy specimen and was described as a white plaque on the subglottic compartment of the left side of the larynx.[5] The case presented here represents the first report of laryngeal glycogenic acanthosis found in a living patient, with a unique presentation as vocal fold leukoplakia.

CASE REPORT

A 49-year-old woman presented to the laryngology clinic for evaluation of a raspy voice of 4–5 weeks duration. She described the onset of the dysphonia as gradual and also reported mild shortness of breath. Her history was significant for former smoking status and current e-cigarette use, as well as intermittent acid reflux.

Videostroboscopy of the larynx was performed in clinic and revealed a white verrucous mass on left true vocal fold free margin (Supporting Video 1). Microlaryngoscopy of the lesion under anesthesia revealed a well-circumscribed, firm, verrucous white lesion involving 80% of the medial aspect of the left true vocal fold, extending nearly to the anterior commissure, but not involving it (Fig. 1). The deep aspect of the lesion appeared to abut the vocal ligament at a small area in the midmembranous portion. A complete excision without additional margins was performed using CO_2 laser to ensure an adequate specimen. While most of the lesion was readily separable from the underlying tissue, the deep extent at the midportion of the lesion was somewhat adherent to the vocal ligament and had to be dissected carefully, raising concern for a possible invasive process.

The histologic sections showed markedly thickened laryngeal squamous mucosa with a flat epithelial-stroma interface and orderly squamous epithelial cell maturation (Fig. 2). There was no evidence of increased mitosis or significant nuclear pleomorphism. The majority of the squamous epithelial cells in the superficial aspects of the surface lining had pinpoint nuclei and prominent cytoplasmic clearing. A Grocott methenamine silver stain to assess for fungal microorganisms was ordered and interpreted as negative. Similar stains with the Periodic acid-Schiff (PAS) method, with and without diastase predigestion, were obtained on separately cut slides of tissue. PAS stains highlighted the cytoplasm of the squamous epithelial cells, while predigestion with diastase abrogated labeling, indicating glycogen as the source of the cytoplasmic clearing. No further pathologic features were

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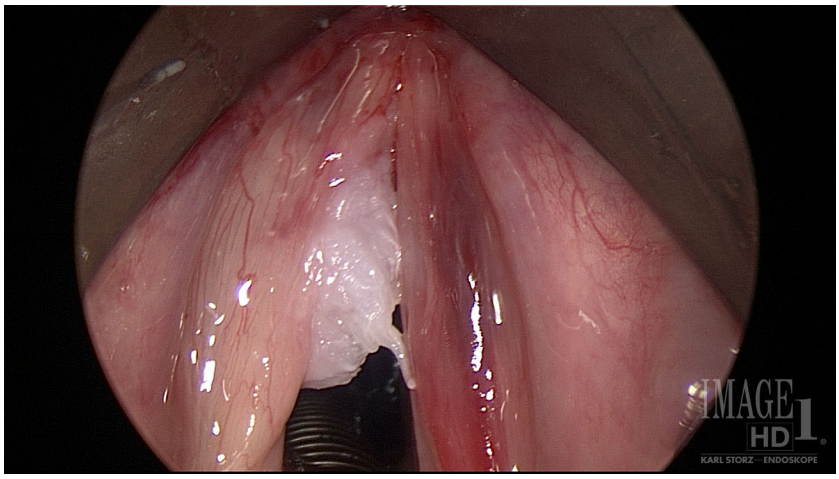

Fig. 1. Intraoperative photograph from microlaryngoscopy demonstrating leukoplakic lesion on left true vocal fold. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

found in multiple additional, deeper level sections. The pathologic diagnosis was determined to be laryngeal glycogenic acanthosis.

DISCUSSION

To our knowledge, this is the first case of glycogenic acanthosis described on the true vocal fold of a living

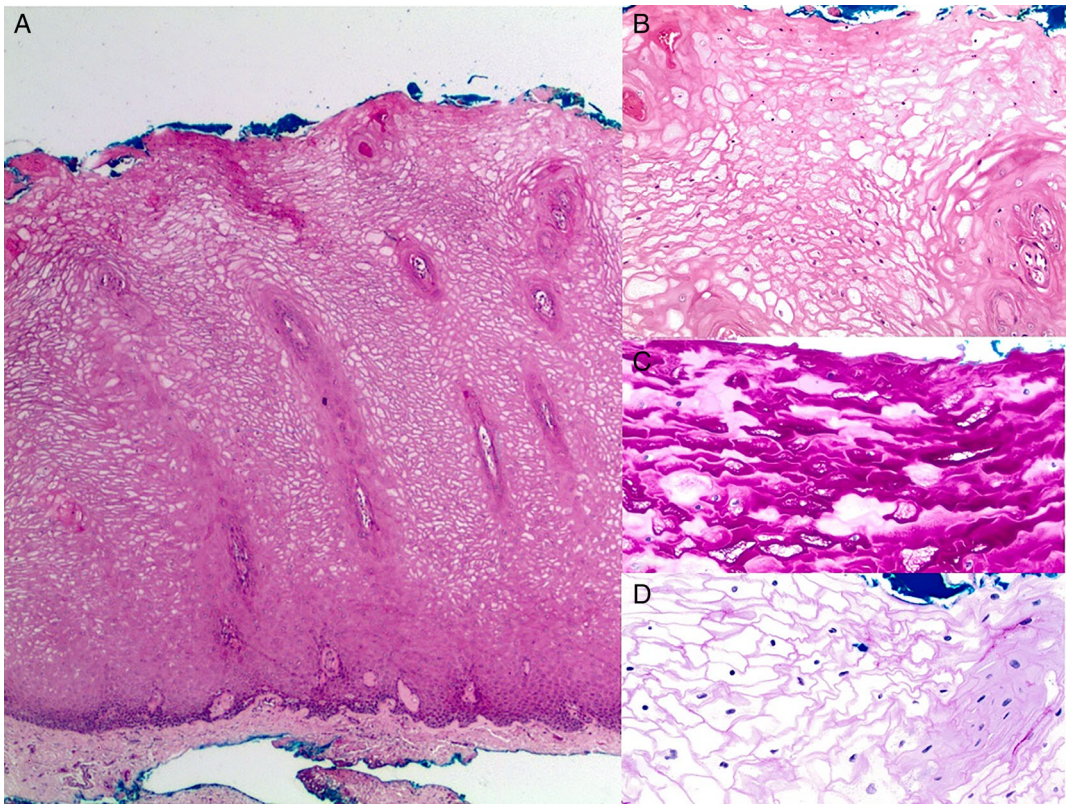

Fig. 2. Histologic slides demonstrating glycogenic acanthosis. (A) Squamous mucosa of larynx, at true vocal cord, low power (hematoxylin and eosin stained). There is thickened squamous epithelium with orderly maturation of squamous epithelial cells. The luminal two-thirds of the squamous epithelium are remarkable for prominent cytoplasmic clearing. (B) Squamous mucosa of larynx, at true vocal cord, intermediate power (hematoxylin and eosin stained). At higher magnification of the luminal third of the squamous epithelium, cytoplasmic clearing is again seen. The nuclei are small and hyperchromatic without evidence of significant pleomorphism or mitotic activity. (C) Squamous mucosa of larynx, at true vocal cord, high power (Periodic acid-Schiff [PAS] method). The cytoplasm contains PAS-positive material. (D) Squamous mucosa of larynx, at true vocal cord, low power (PAS method after diastase pretreatment [PASD]). The PAS-positive material is no longer present after pretreatment with diastase, confirming that the PAS-positive material in panel (C) is glycogen. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

patient. Although the clinical implication of this diagnosis is unclear, its associations with GERD and insulin resistance in the gastroenterology literature could lead to speculation about similar associations when involving the larynx. This patient did report a history of intermittent acid reflux symptoms. She was also mildly obese. However, hemoglobin A1c at the time of diagnosis was not elevated. Following excision of the lesion, the patient reported improvement in her voice, and no recurrence of the lesion was noted at the time of her most recent follow-up visit at 6 months.

CONCLUSION

Ultimately, the significance of this pathology with regard to systemic disease is undetermined, and further investigation may support an association with laryngopharyngeal reflux disease. However, it is important to now recognize glycogenic acanthosis as a potential benign etiology that can present as a vocal fold mass with leukoplakia.

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