“Papillomatoma”: A Rare Case of Subepithelial Papilloma of the Vocal Fold

Jeffrey P Marino1* and Adam M Klein2

1Department of Otorhinolaryngology and Communication Sciences, Ochsner Medical Center, USA
2Department of Otolaryngology-Head and Neck Surgery, Emory University School of Medicine, USA

Abstract
Laryngeal papillomatosis is a recurrent condition typically manifesting as benign epithelial lesions affecting glottic closure and vocal fold mucosal wave propagation. This case describes a subepithelial deposit of papilloma which developed as a complication of prior surgical interventions that disrupted the vocal fold epithelium and violated its microarchitecture.

ABBREVIATIONS
LP: Laryngeal Papillomatosis; HPV: Human Papilloma Virus; PDL: Pulsed Dye Laser; KTP: potassium-Titanyl-Phosphate; VRQOL: Voice Related Quality of Life

INTRODUCTION
Laryngeal papillomatosis (LP) is characterized by the distribution of papilloma along the surface epithelium and mucosa of the larynx. Consequences of these lesions range from mild dysphonia to severe respiratory distress necessitating emergency airway intervention. The causative agent is the human papilloma virus (HPV), with types 6 and 11 responsible for most cases but other serotypes also reported [1,2]. The mainstay of treatment is surgical intervention, utilizing techniques such as microsurgical excision [3], carbon dioxide laser excision [4], cryotherapy [5], microdebrider removal [6], and photoangiolyis with either the 585 nm pulsed dye laser (PDL) [7] or the pulsed 532 nm potassium-titanyl-phosphate (KTP) laser [8]. Occasionally, local injections of cidofovir [9,10] or bevacizumab [11] are implemented for more aggressive disease. The goals of surgery are to optimize airway and preserve voice by avoiding damage to the subepithelial, non-diseased tissue so as to prevent adverse laryngeal sequelae including scarring, stenosis, or deeper seeding of disease. The contemporary management of LP may incorporate unsedated office-based treatments [12] and additional imaging modalities [13,14]. A potential role for the HPV vaccine to limit recurrence is being investigated [15].

Here we report a case of a subepithelial vocal fold inclusion cyst comprised of laryngeal papillomatosis, or a “papillomatoma,” which is suspected to have developed as a complication of prior surgical debridements.

CASE PRESENTATION
A 42-year-old male preacher presented for further management of a longstanding diagnosis of adult-onset recurrent respiratory papillomatosis. The patient reported multiple prior surgical procedures in the operating room at outside institutions. He complained of vocal roughness, increased vocal effort, decreased vocal endurance, and increased vocal fatigue. Voice-related quality of life (VRQOL) index raw score was 46. His voice was characterized by moderate roughness and strain, with irregular, mild phonatory breaks. The remainder of his history and routine physical examination were unremarkable. Rigid 70-degree laryngeal videostroboscopy (Pentax, Tokyo, Japan) identified bulky lesions along the entire left true vocal fold, as well as a small nidus of disease on the mid-right true vocal fold, consistent with laryngeal papillomatosis. The recommendation was made for treatment of disease in the operating room. The patient was brought for suspension microlaryngoscopy with biopsy and photoablation of the papilloma with the 532 nm pulsed KTP laser (Boston Scientific, Natick, MA). Findings were consistent with those identified in the office but also included a small anterior glottic web, a mild loss of superficial lamina propria on the left vocal fold, and a small subepithelial cystic lesion in the anterior third of the left true vocal fold (Figure 1), which was left unaddressed at this time. The patient underwent routine surveillance of disease over the next three years, with minor progression of disease that was managed in the awake setting using the pulsed KTP laser on four occasions.

Over the course of the patient’s treatment, while the surface papilloma responded well to KTP laser treatments, the left vocal fold subepithelial lesion began to grow in size such that it was negatively impacting the patient’s voice by interfering with glottic closure and vocal fold vibration. As a consequence, the patient developed moderate roughness and instability in his vocal quality. Eventually the patient was brought back to the operating room for suspension microlaryngoscopy. After superficial disease was treated with the pulsed KTP laser, a laryngeal sickle blade was used to make an epithelial cordotomy in the left true vocal fold, lateral to the subepithelial lesion. Careful dissection with microflap preservation identified a subepithelial inclusion of papilloma encapsulated within dense scar (Figure 2). Biopsy of the contents of the lesion was consistent with benign laryngeal papillomatosis. The papilloma was debulked from within the capsule using cold instrumentation, followed by pulsed KTP laser photoablation.

No further procedures have been necessary at the time of this publication. At the patient’s most recent follow-up visit, over two years since surgery, the patient reported significant improvement in his voice, with a VRQOL raw score of 10. His voice was characterized by slight roughness but otherwise demonstrated marked improvements in strength and stability. Videostroboscopy demonstrated complete resolution of the left vocal fold “papillomatoma” with only minor recurrence of superficial papilloma along the right and posterior left vocal folds.

**DISCUSSION**

The most commonly cited complications of LP surgery include anterior glottic webbing, vocal fold scarring, and laryngeal stenosis [16-21]. These complications can be avoided by treating only the involved epithelium and preserving the integrity of the underlying superficial lamina propria. While multiple surgical technologies are available for treatment of the disease, a recent review noted a trend toward increased utilization of microdebrider [22]. Regardless of which technology is employed, overaggressive surgery can result in disruption of the epithelium, as well as the deeper layers of the vocal fold microarchitecture. When the virus is seeded in the submucosal vocal fold, presumably with a nidus of epithelium, papilloma may then proliferate in atypical locations.

To date, there have been no reports in the literature of a deposit of papilloma in a location beneath the epithelium of the vocal fold. This case report highlights a unique potential complication of LP surgery that can pose challenges for both surveillance and treatment.

**REFERENCES**


