Consecutive presentations of laryngeal oncocytic cystadenomas

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ABSTRACT

Objectives: To present three cases of laryngeal oncocytic cystadenomas and summarize current recommendations for treatment.

Methods: A retrospective chart review of three patients with oncocytic laryngeal cystadenomas, treated with primary surgical intervention at a tertiary care center, was performed.

Results: Three patients, two female and one male, with mean age 68, two presented with progressive dysphonia and globus, and one presented with otalgia. Physical examination revealed a cystic structure arising from the right posterior vestibule in one, a fullness of the right aryepiglottic fold in another, and fullness of the right vallecula in the final patient. Each patient underwent endoscopic CO2 laser assisted resection of the laryngeal mass. In all three cases, pathological analysis revealed oncocytic cystadenoma, with clear margins.

INTRODUCTION

Laryngeal cysts may be classified as neoplastic or nonneoplastic cysts. Neoplastic cysts include congenital cysts, traumatic cysts, lymphangiomas, and hemangiomas. Nonneoplastic cysts include glanular cysts, amylodaiat cysts, and lymphatic cysts. Glanular cysts, also termed cystadenomas, papillary cystadenomas, oncocytic adenomas, or oncocytic cystadenomas, are rare lesions whose epithelial components resemble that of Warthin's tumors found in the major salivary glands. They are typically considered separately from solid adenomas of the larynx. Although imaging characteristics of cysts were reported as early as 1891, the first case series, comprising four such cases, was reported in 1944. The most common laryngeal site affected is the supraglottis, and lesions usually present in the seventh or eighth decades of life. They are occasionally diffuse or multilocal but more commonly present as single lesions causing hoarseness. If large enough, they may cause upper airway obstruction.

While many agree that the etiology is primarily neoplastic, delayed hypersensitivity has been cited as an inciting factor, similar to what has been suggested for Warthin's tumors. Optimal treatment involves surgical removal, usually endoscopic, although larger or recurrent tumors may require open laryngeal surgery.

Here we present a series of three cases of laryngeal oncocytic cystadenoma, each treated successfully with endoscopic resection.

METHODS

This study was exempt from institutional review board approval at the University of California, Los Angeles. Inclusion criteria included a histologic diagnosis of oncocytic cystadenoma involving any portion of the larynx from 2013 to 2014. Three patients were identified, and their medical records were analyzed for demographic and clinical data.

RESULTS

There were two females and one male, with an average age at presentation of 68 (range, 65 to 70). In two patients, presenting symptoms included globus and dysphonia; in the third, the only presenting symptom was unilateral otalgia. One patient denied ever having smoked; the remaining two patients reported prior tobacco use of 25-pack-years and 50-pack-years, respectively.

Preoperative videolaryngoscopy was performed in all three patients. In one patient, only a fullness of the left aryepiglottic fold was noted. In the second, a cyst was noted in the posterior aspect of the right vestibule. In the third, a cystic mass obstructing the anterior aspect of the right vestibule was noted. Cross-sectional imaging was performed in all three patients; the magnetic resonance imaging findings of the third patient described above are shown in Figure 1.

Each patient underwent operative microsuspension direct laryngoscopy under general anesthesia for resection of the laryngeal cyst (Figure 2). In each case, a carbon dioxide line-of-sight laser was used for microdissection. In all three cases, the cyst was removed en bloc (Figure 3). Following resection of the cysts, exposed mucosal surfaces were allowed to heal by secondary intention.

CONCLUSIONS

Oncocytes are cells with eosinophilic, granular cytoplasm following hematoxylin-eosin staining. They are found in a variety of lesions presenting in the head and neck, including oncocytes, Warthin tumors, salivary duct carcinomas, Hurthle cell lesions, granular cell tumors, parangangiomas, and melanomas. In particular, oncocyctic cystadenomas have been described in the larynx, predominantly affecting elderly patients, and are typically cystic or complex on imaging. Clinical presentation and imaging may be consistent with laryngocoele, and a conservative approach at complete excision is both diagnostic and therapeutic. If clear margins are obtained, no adjuvant therapy is indicated. Although laryngocoeles and malignancies are more commonly seen in this population, oncocytic cystadenomas should remain in the differential of a cystic or complex laryngeal mass.

Figure 4. Histologic appearance of laryngeal oncocytic cystadenoma. The supraglottic cystic lesion is lined by monolayer or haphazardly-arranged oncocytic cells. These oncocytic cells have small, round, uniform nuclei and abundant eosinophilic granular cytoplasm. Focally papillary projections are also seen. Bilayered oncocyes and lymphoid stroma as seen in Warthin's tumor are absent.

Pathological analysis revealed an oncocytic cystadenoma in all three cases (Figure 4). Specifically, the cystic lesions were noted to be lined by monolayer or haphazardly-arranged oncocytic cells. These oncocytic cells had small, round, uniform nuclei and abundant eosinophilic granular cytoplasm. Focally papillary projections were also seen. Bilayered oncocyes and lymphoid stroma as seen in Warthin's tumor were absent.

Median follow-up was 6.5 months (range, 6 to 7 months); one patient who presented initially with globus and dysphonia was lost to follow-up postoperatively. At most recent follow-up, the remaining two patients were free of their presenting symptoms, and on laryngeal videostroboscopy, bilateral mucosal waves were noted to be present and symmetric.

DISCUSSION

Laryngeal oncocytic cystadenomas, which can resemble Warthin's tumors of the major salivary glands on histological analysis, are rare, benign neoplasms found most commonly in the supraglottis. Numerous terms, including oncocytic adenoma, eosinophilic granular cell cyst, oncocytoma, oncocyctic cyst, and oncocytic papillary cystadenoma, have all been used to describe the same entity. They can mimic other cystic laryngeal pathologies, including laryngocoele and congenital laryngeal cysts. The most common presenting symptoms are dysphonia and globus, although otalgia may be the only presenting complaint, as evidenced one of the patients in this series.

The pathogenesis of the lesion remains under debate. While most believe that the lesion is a true neoplasm, some believe that the lesion is reactive in nature, manifesting as the end result of metaplasia from the ductal epithelium of mucous glands found in the larynx.

Treatment is primarily surgical. Most laryngeal oncocytic cystadenomas are amenable to endoscopic excision with clear margins, although open laryngeal procedures may be considered if disease is extensive or recurrent. No adjuvant therapy is typically given. Follow-up with endoscopy at regular intervals is indicated to surveil for recurrence. The otolaryngologist must remain cognizant of this entity, as it can closely mimic laryngocoeles in presentation as well as endoscopic and cross-sectional appearance.