Title
Acute dyspnea caused by a giant spindle cell lipoma of the larynx

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CASE REPORTS

Spindle Cell Lipoma Arising from the Supraglottis: A Case Report and Review of the Literature

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Abstract
Lipomas are common benign mesenchymal neoplasms. Although 13% of lipomas are found in the head and neck, only 0.6% have been reported in the larynx. Of all lipomas, the spindle cell variant is the least common. In the present study, we report a case of supraglottic spindle cell lipoma and review the literature of laryngeal spindle cell lipoma. A 35-year-old male presented with dysphagia and dyspnea and was found to have bilateral supraglottic lesions causing airway obstruction. The masses were resected endoscopically. Final pathology demonstrated mature adipocytes and spindle cells, with immunohistochemical patterns supportive of spindle cell lipoma. Spindle cell lipomas have rarely been reported in the upper airway. To our knowledge, this is the youngest patient reported to date. These lipomas are uncommon benign neoplasms and should be distinguished from aggressive mesenchymal neoplasms such as liposarcoma variants to guide appropriate conservative but curative therapy.

Keywords Spindle cell lipoma · Supraglottis · Laryngeal mass · Dyspnea · Head and neck lipoma

Introduction

Lipomas are a group of adipocytic mesenchymal neoplasms that frequently arise in the head and neck. They are most commonly found in the posterior neck and represent only 0.6% of benign laryngeal tumors [1, 2]. Most such tumors are easily distinguishable histologically. The spindle cell variant, which is composed of mature adipocytes and bland spindle cells, has been rarely identified in the larynx and hypopharynx [3]. In the present report, we discuss a case of spindle cell lipoma arising from the supraglottis and extending into the larynx of a 35-year-old male. We review the literature of laryngeal spindle cell lipoma with emphasis on diagnosis and proper therapeutic management.

Case Report

A 35-year-old male underwent tonsillectomy and septoplasty for obstructive sleep apnea at an outside institution. At the time of intubation, he was found to have a mass originating from the left arytenoid, and excisional biopsy was interpreted as laryngeal chondroma. He was lost to follow up until he developed dysphagia and dyspnea three years later. A contrast-enhanced neck CT demonstrated a 3.7 cm heterogeneous lesion with internal septations and fat arising from the left hypopharynx extending into the left pyriform sinus (Fig. 1). The patient was referred to the senior author (EA) for further evaluation.

In the operating room, the patient underwent tracheostomy followed by direct laryngoscopy, which revealed a left supraglottic mass encroaching on the left pharynx as well as a separate right supraglottic mass. Both masses were excised endoscopically with CO_2 laser. He was subsequently decannulated prior to discharge with relief of airway difficulties and dysphagia.

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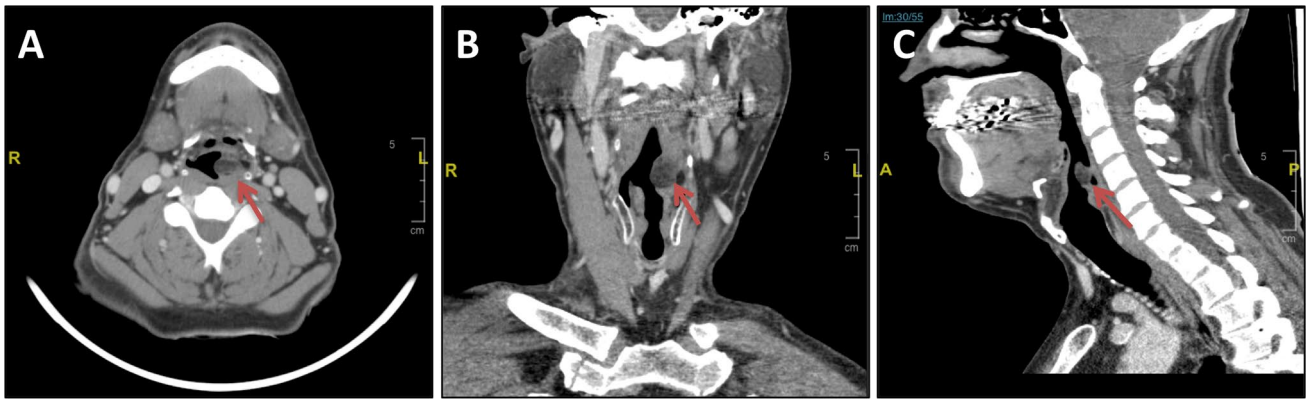

Fig. 1 CT neck with contrast depicting the supraglottic mass in **a** axial, **b** coronal, and **c** sagittal views. The red arrow indicates the mass.

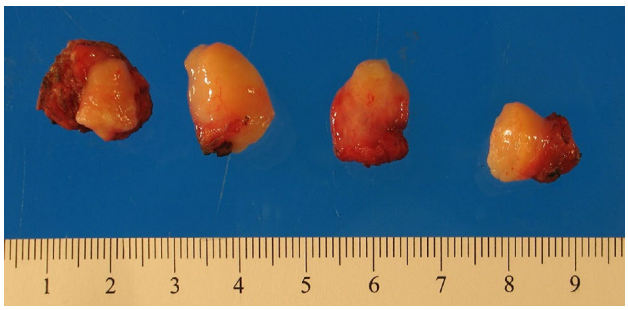

Fig. 2 Gross photograph of left vocal cord mass, serially sectioned.

Gross pathology of the lesions demonstrated soft, ovoid masses with red-tan to white-tan cut surfaces (Fig. 2). The left supraglottic lesion measured 3.0 cm by 2.0 cm by 1.5 cm; the right lesion measured 2.0 cm by 1.5 cm by 1.5 cm. Microscopic examination showed a fairly well-circumscribed mass underlying unremarkable squamous mucosa (Fig. 3a), consisting of mature adipocytes and uniform, bland spindle cells arranged in collagen-rich fascicles with occasional thick ropey collagen strands. Adipocytes and stroma were present in three patterns: (1) an admixture of adipocytes with fibrous cords (predominant pattern; Fig. 3b), (2) solid fibrous areas (Fig. 3c), and (3) purely lipomatous areas (the least frequent pattern; Fig. 3a). Regardless of pattern, the stromal cells stained uniformly positive for CD34 and negative for S100 (Fig. 3d and data not shown); more than 50% of stromal cells demonstrated loss of the phosphorylated retinoblastoma protein (pRb) (Fig. 3e). The adipocytes were uniformly mature and stained positively for S100 (data not shown). These features together are diagnostic of a spindle cell lipoma [3]. Given these benign findings, no further treatments were recommended.

Discussion

Laryngeal lipomas are uncommon; a review of PubMed reveals fewer than 150 cases reported in the English literature. They comprise only 0.6% of benign laryngeal tumors and are more prevalent in men [1, 3]. Of these cases, six reported spindle cell lipoma variant, as summarized in Table 1. Laryngeal spindle cell lipomas are also more common in men and the average age at presentation is 60.3 years old. Clinical presentations vary but usually include dyspnea, dysphagia, and globus sensation.

The first reported case of spindle cell lipoma in the larynx was by Nonaka et al. in 1993, in which a 2.6 cm spindle cell lipoma was identified in the right vestibular fold and ventricle, and excised via a laryngofissure approach [4]. Since then, six other cases of spindle cell lipoma of the larynx have been reported, all of which were removed endoscopically. Lee et al. excised a hypopharyngeal spindle cell lipoma from the pyriform sinus using transoral robotic surgery and described the benefits of this technique, namely the uninhibited lateral and inferior visualization, for fully resecting such hypopharyngeal masses [5]. This approach could be considered similarly for a laryngeal tumor. In our case, adequate visualization was achieved with microdirect laryngoscopy and full excision was possible with in-sight and handheld CO_2 laser. The rarity of this tumor precludes a formal comparison of recurrence rates using either technique. However, it is clear that full visualization of the tumor and its pedicled site is needed for adequate excision using either cautery or laser tools.

It is important to distinguish these tumors from other neoplasms because overtreatment can result from incorrect assessment of their benign nature. Spindle cell lipomas can be mistaken for well-differentiated liposarcomas/atypical spindle cell lipomatous tumors (WDLS), especially when

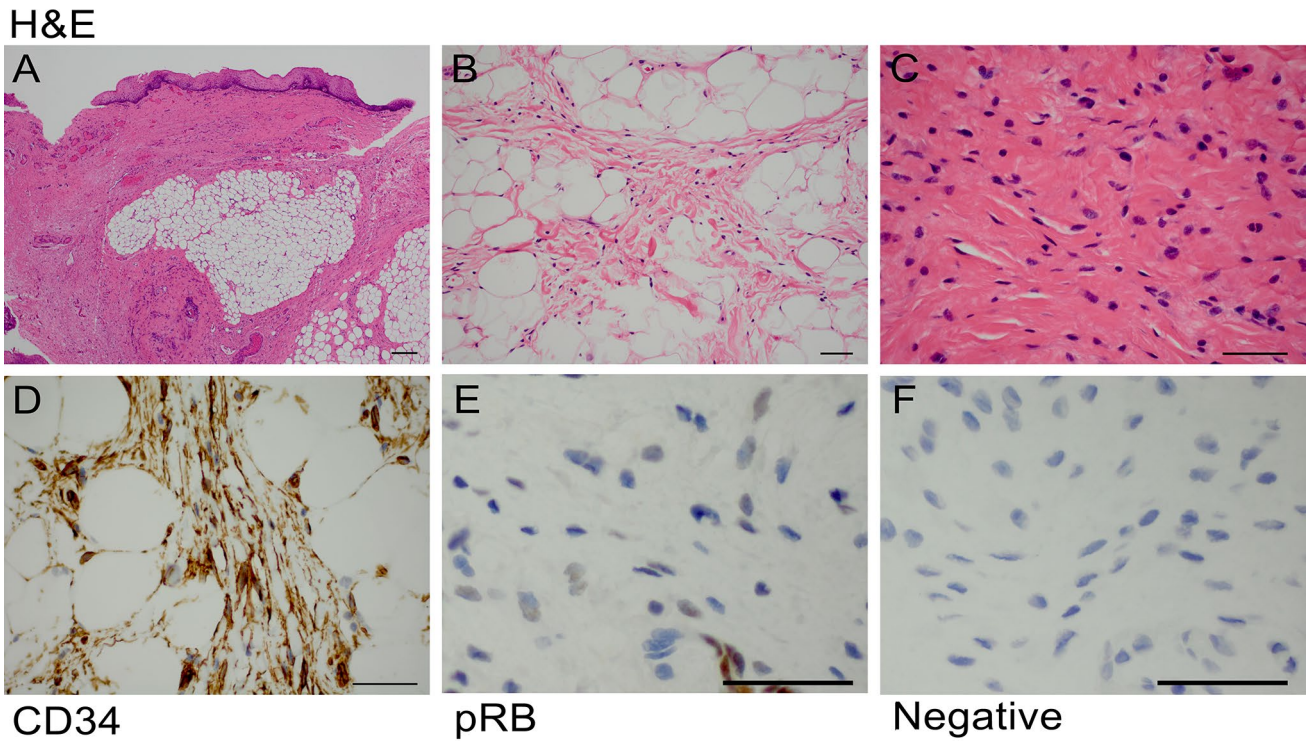

Fig. 3 Microscopic exam of left vocal cord mass. **a–c** Hematoxylin and eosin (H&E) stained section of the left vocal cord mass demonstrating the **a** sub-mucosal location and **b**, **c** heterogeneity in adipose and solid fibrous patterns. **d** CD34 immunostain demonstrating uniform stromal cell staining. **e** pRB immunostain demonstrating loss of nuclear staining in greater than 50% of stromal cells. Blood vessel on bottom right shows intact staining. **f** Negative control. Scale bar in **a** represents 200 micrometers and in **b–f** represents 50 micrometers.

Table 1 This is a summary of case reports of laryngeal spindle cell lipoma.

Author	Presentation	Location	Treatment
Nonaka et al., 1993 [4]	68-year-old male with a six month history of dysphonia	2.6 cm mass in the right vestibular fold and ventricle	Laryngofissure
Nader et al., 2012 [9]	63-year-old male with a several month history of snoring and dyspnea	2.7 cm mass that was pedicled at the left aryepiglottic fold that ball-valved into the glottis causing partial obstruction	Endoscopic
D'Antonio et al., 2013 [10]	65-year-old male with a two week history of dysphonia, dyspnea and stridor	Large polyp of the left true vocal fold with a wide pedicle with a smooth surface that ball-valved in the glottis	MDL[a]
Kodiyan et al., 2015 [11]	79-year-old female with dysphonia for one year, globus sensation	3.5 cm submucosal mass in the right supraglottis from the arytenoid to the aryepiglottic fold	MDL and laser
Wolf-Magele et al., 2016 [12]	52-year-old male who presented with neck pain, stridor, and dyspnea	8×5 cm laryngeal mass	CO2 laser
Azar et al., 2020	35-year-old male with progressive dyspnea	4 cm mass on the left lateral pharyngeal wall, pyriform sinus and supraglottis. 2 cm right supraglottic mass	Tracheostomy, MDL and laser

[a] Microdirect laryngoscopy

found in a deep location such as the larynx. Other lipomatous or spindle cell lesions such as schwannoma, neurofibroma, angiolipoma, dermatofibrosarcoma protuberans, and solitary fibrous tumors, share histologic similarities and may also be considered in the differential diagnosis [6].

Histologic evaluation and cytogenetic studies are useful tools for differentiating these lesions in order to guide

appropriate management. Spindle cell lipomas are composed of uniform spindle cells, thick collagen fibers, and variably prominent mature adipocytes. They result from a loss of expression of tumor suppressor pRb, which can be visualized by immunohistochemistry [3]. WDLS are more aggressive neoplasms, often arising from deep mesenchymal tissues; as these may show histologic overlap with spindle cell lipomas, they should be considered in the differential diagnosis. They show enlarged, hyperchromatic, pleomorphic cells and often show variability in adipocyte size [7]. While both tumors may contain CD34-expressing bland spindle cells, WDLS have intact pRb expression, and show MDM2 gene amplification by cytogenetic analysis [8]. In this case, the characteristic histology along with the loss of pRb were most consistent with a diagnosis of spindle cell lipoma.

The present case has a few unique features that contribute to our current understanding of laryngeal spindle cell lipoma. While these benign neoplasms have been noted to usually affect those of older age, here we present the youngest known case at 35 years old. Another notable feature of the spindle cell lipoma described here is its presentation as two separate bilateral masses in the supraglottic space. In the current literature, there are no other reports of multiple, bilateral spindle cell lipomas in the larynx. Moreover, it is important to underline the careful histologic evaluation of this tumor in order to prevent over-treatment. Initially, our patient's tumor was interpreted as a chondroma. Had this been confirmed, a wider margin-free resection would have been appropriate. In contrast, with the diagnosis of a non-malignant lipoma, simple removal of the tumor and its pedicle was appropriate.

Conclusions

The present case report describes a 35-year-old male with bilateral supraglottic spindle cell lipomas. Lipomas of the larynx are uncommon benign neoplasms, and spindle cell variant is even more rare. However, spindle cell lipoma should be considered in the differential diagnosis of fibrous or fatty lesions of the larynx and hypopharynx and should be distinguished from more aggressive mesenchymal neoplasms to guide appropriate treatment.

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Compliance with Ethical Standards

Conflicts of interest The authors have no conflicts of interest to declare that are relevant to the content of this article.

IRB Approval This study was deemed exempt by the Institutional Review Board at the University of California, Los Angeles due to the de-identified nature of the report.

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