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What is This?
Primary Laryngotracheal Melanoma

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Keywords
laryngeal melanoma, tracheal melanoma, mucosal melanoma, primary, head and neck

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Introduction
Head and neck mucosal melanomas represent 0.7% to 3.8% of all melanomas.¹ Among this group, primary melanoma of the larynx and trachea are very rare. There are 8 reports of primary tracheal melanoma and about 60 reported cases of primary laryngeal melanoma in the literature. We present an unusual case of primary laryngotracheal melanoma. This article was submitted for review by our institutional review board and deemed not subject to review.

Case Presentation
A 64-year-old African American man initially presented with 4 to 5 months of progressive dyspnea on exertion, cough productive of gray/black material, hoarseness, dysphagia, and a 10- to 15-lb weight loss. Medical history was notable for chronic obstructive pulmonary disease but negative for hemoptysis, asthma, chest pain, or other systemic illnesses, and he had not had any prior cutaneous lesions. Social history was notable for a 21 pack-year smoking history. On examination, he had stridor, and fiber-optic laryngoscopy revealed a pigmented, charcoal lesion involving the aryepiglottic folds, arytenoids, circumferential false vocal folds, glottis, and subglottis (see Video 1, available at otojournal.org).

Computerized tomography (CT) of the neck revealed a mass of approximately 1.3 × 4.9 cm in the larynx and subglottic airway, causing significant focal subglottic airway narrowing (see Figure 1). Direct laryngoscopy and biopsy by the referring physician revealed malignant melanoma, which stained positive for pan melanin and negative for cytokeratin and S100. His stridor worsened, and he was taken to the operating room for awake tracheostomy under local anesthesia, as well as bronchoscopy, direct laryngoscopy, esophagoscopy, and gastrostomy. His melanoma extended nearly to the carina with progression of obstruction at the glottis (see Figure 2). No synchronous lesions were identified. Staging positron emission tomography (PET)/CT showed abnormal uptake at the laryngotracheal mass without metastasis. He underwent radiation therapy, receiving 74 Gy in 37 fractions over 7 weeks. The first posttreatment PET/CT showed partial interval response to therapy with reduction in abnormal fludeoxyglucose uptake corresponding to the previously noted laryngotracheal mass. Subsequently, fiber-optic examination demonstrated partial treatment response with remnant striations of charcoal pigment still evident. New nasal mass was viewed in the right vestibule (see Video 2, available at otojournal.org). Three-month posttreatment PET/CT demonstrated an interval of remarkable progression of the tumor in the larynx, pharynx, and trachea, with additional new nasal mass, innumerable lung metastases, hilar tumor, cervical intraspinal muscular tumor, and suspected progressive liver tumor. He was then treated with ipilimumab chemotherapy but had gross progression of disease during treatment and eventually entered hospice care 10 months from diagnosis.

Discussion
Epidemiology
Primary laryngotracheal melanoma is extremely uncommon. Gaissert et al² performed a retrospective review of 360

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primary tracheal tumors, in which only 1 patient was identified with tracheal melanoma. In a review of 259 cases of head and neck mucosal melanoma, the most common mucosal sites were the nasal cavity (69%) and the oral cavity (22%).3 Of the reported cases of primary laryngeal melanoma in the literature, Terada et al4 reviewed 38 patients, with sufficiently detailed reports available, and found 80% were male, the mean age was 60 years, and the supraglottis was the most common primary site.

Diagnosis

The diagnosis of a head and neck mucosal melanoma is established through biopsy and pathologic examination. Criteria have been proposed for the differentiation of primary versus metastatic melanoma of the larynx and trachea.5

Prognosis

Of the laryngeal melanoma cases reviewed by Terada et al,4 2 of 28 cases survived for more than 5 years, and the overall 3- and 5-year survival rates were 28.6% and 7.1%, respectively. In addition, most of the cases they reviewed had regional lymph node metastasis, and most had distant metastasis.4 Most primary tracheal melanoma cases report less than 1 year survival from diagnosis.

Treatment

Surgical excision is required for primary lesions when possible; treatment for metastatic melanomas is palliative. Radiotherapy has been used with limited success as a single modality therapy for laryngeal melanoma; it may be useful as a surgical adjuvant and in the primary management of unresectable disease.5 Newly introduced systemic therapies such as vemurafenib and ipilimumab used in metastatic melanoma may prove to be of benefit in some patients with metastatic laryngotracheal melanoma. The prognosis for laryngotracheal melanoma remains poor irrespective of the treatment modalities used.

Author Contributions

Jonathan Mark, (1) substantial contributions to conception and design, acquisition of data, and analysis and interpretation of data; (2) drafted the article and revised it critically for important intellectual content; and (3) final approval of the version to be published; Salvatore Taliercio, (1) substantial contributions to conception and design, analysis and interpretation of data; (2) revised the article critically for important intellectual content; and (3) final approval of the version to be published; Daniel Karakla, (1) substantial contributions to conception and design, analysis and interpretation of data; (2) revised the article critically for important intellectual content; and (3) final approval of the version to be published.

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References


