Oncocytic Glomus Tumor of the Trachea*

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An oncocytic variant of glomus tumor of the trachea occurred in a 47-year-old woman. She experienced intermittent cough and hemoptysis for about three years. Bronchoscopy and chest CT scan showed a small reddish polypoid tumor on the lower end of the trachea. Bronchoscopic biopsy was carefully done and was diagnosed as oncocytoma. A wedge resection of the tumor was done. Tumor cells were characterized by strongly eosinophilic granular cytoplasm on light microscopy and by numerous closely packed round or ovoid mitochondria with prominent tubular cristae on electron microscopy. They were arranged in a sheet around small vessels, as a result of which the biopsy diagnosis of oncocytoma was changed to oncocytic glomus tumor. To our knowledge, this is the first report of an oncocytic glomus tumor arising from the trachea.

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Glomus tumors are uncommon benign neoplasms which are commonly seen in the extremities, particularly in the nail bed and finger tip of young adults. This tumor was first described by Masson1 in 1924. Since then, sporadic reports of glomus tumors of varying types and locations have appeared in the literature. According to the relative proportions of glomus cells, vascular structures, and smooth muscle tissue, glomus tumors have been divided into three subtypes, namely: classic glomus tumor, glomangioma, and glomangiomyoma.2 The trachea is a very unusual site for this tumor, and only four cases of tracheal glomus tumors have been reported in the literature.3,4

Recently, oncocytic glomus tumor, a new variant, was described.5 This was characterized ultrastructurally by numerous mitochondria resulting in oncocytic change on light microscopy, which was distinct from the three subtypes of glomus tumor.

We report a case of tracheal glomus tumor characterized by oncocytic change on light microscopy and numerous close-packed mitochondria on electron microscopy. We believe that this is the first case report of oncocytic glomus tumor of tracheal origin.

CASE REPORT

A 47-year-old woman was admitted to Hanyang University Hospital in February 1989 because of intermittent cough and hemoptysis. She had been well until three years ago, when her symptoms occurred infrequently. Two years later, the hemoptysis became somewhat aggravated. The findings from physical exami*

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FIGURE 1. Bronchoscopy shows small reddish mass on trachea just above carina, which is slightly bleeding by touch.

nation were normal except for arrhythmia. The chest x-ray film and results of pulmonary function tests were normal. The ECG showed a few PVCs. Fiberoptic bronchoscopy revealed a small reddish polypoid mass protruding into the lumen of the trachea just above the carina (Fig 1). Bronchoscopic biopsy was obtained carefully, and the bleeding during biopsy was not significant. A chest CT scan showed a small nodular mass on the right posterolateral portion of the lower end of the trachea, without evidence of regional invasion. Under the diagnosis of oncocytoma, the patient underwent a wedge resection of tumor, and direct closure was performed. The patient was discharged in March 1989 and has been followed at the outpatient clinic without problems.

The wedge resection of trachea contained a 1.5 × 1 × 1-cm multilobulated polypoid mass. The lobulated mass was reddish brown and appeared to be arising from the mucosa.

The light-microscopic features of the resected specimen were identical to those of biopsy specimen. A well-demarcated mass occupied the subepithelial connective tissue and smooth muscle layer of the trachea. The covering epithelium was intact. The tumor was characterized by organoid nests and solid sheets of uniform round or polygonal cells around the thin-walled vascular spaces.

FIGURE 2. Tumor cells show eosinophilic granular cytoplasm (hematoxylin-eosin, original magnification × 400).
The tumor cells had eosinophilic granular cytoplasm with well-defined cell borders (Fig 2). The nuclei were round to oval, with finely dispersed chromatin, and rare mitotic figures were present. The tumor was highly vascular with thin-walled interlacing vessels, some of which were dilated. The reticulin stain revealed many fine reticulin fibers lying between individual tumor cells. No intracytoplasmic granules of the carcinoid type were stainable by the Grimelius method. The histologic features of the tumor represented an oncocytic glomus tumor, rather than an oncocytoma of the mucus gland type.

On electron microscopy, the tumor cells had numerous closely packed round or elongated mitochondria with predominant tubular cristae. At the margin of the cytoplasm, there were intracytoplasmic microfilaments with focal densities and dense plaques on the cell membrane. Many tumor cells were surrounded by a basal lamina. There were occasional pinocytotic vesicles along the plasma membrane, and small amounts of glycogen, free ribosomes, and smooth and rough endoplasmic reticula were present (Fig 3). No membrane-bounded dense core granules or desmosomes were seen. The nuclei were uniform and oval with peripheral condensation of chromatin. Occasionally, a nucleolus and nuclear bodies were also noted. Scattered mast cells were seen. The electron-microscopic findings were consistent with glomus tumor with oncocytic features (oncocytic glomus tumor).

**DISCUSSION**

A primary tracheal tumor is one of the rarest tumors, and it is often diagnosed late in the clinical course and at an advanced stage. The majority of such tumors are either squamous cell carcinoma (45 percent) or adenoid cystic carcinoma (36 percent), and the minority (11.4 percent) are of mesenchymal origin. Complaints are coughing, hemoptysis, and dyspnea with wheezing. Endoscopic examination remains one of the most reliable means of establishing the diagnosis of a tracheal neoplasm, and it should be employed in a patient with a normal chest roentgenogram who has an unexplained cough, hemoptysis, wheeze, or dyspnea.

Glomus tumor is a relatively benign uncommon neoplasm found in all parts of the body, which is most commonly seen in the subungual area. The tumor is a distinctive type of vascular tumor whose cell type is a modified smooth muscle cell closely resembling the glomus body from which the name is derived. Unusual sites of origin have included the stomach, mediastinum, vagina, penis, lung, and trachea. At the ultrastructural level the glomus tumor cells appear to be epithelioid smooth muscle cells characterized by a basement membrane envelope, pinocytotic vesicles along the plasma membrane, and cytoplasmic myofilbrils with focal electron densities. We have found only four cases of tracheal glomus tumor reported in the literature (Table 1).

Recently, Slater et al described a new oncocytic variant of Masson's glomus tumor in a 37-year-old man with a subcutaneous neoplasm located in the left thigh. The tumor cells were characterized ultrastructurally by numerous closely packed round or ovoid mitochondria with predominant tubular cristae resulting in oncocytic (oxyphilic) change. The chicken-wire network of reticulin encircled individual tumor cells. The ultrastructural features were suggestive of smooth muscle differentiation. The latter included basal

**Table 1—Clinical Findings and Histologic Types of Tracheal Glomus Tumors**

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>Age (yr)</th>
<th>Sex</th>
<th>Clinical Features</th>
<th>Location in Trachea</th>
<th>Size, cm</th>
<th>Histologic Type</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hussarek and Riedler</td>
<td>1950</td>
<td>43</td>
<td>F</td>
<td>Dyspnea</td>
<td>Subglottic</td>
<td>2.5 × 1.0</td>
<td>Glomangioma</td>
</tr>
<tr>
<td>Fabich and Hafez</td>
<td>1980</td>
<td>63</td>
<td>M</td>
<td>Airway obstruction</td>
<td>Above bifurcation</td>
<td>2.5 × 1.0</td>
<td>Glomangioma</td>
</tr>
<tr>
<td>Heard et alb</td>
<td>1982</td>
<td>50</td>
<td>M</td>
<td>Asthma-like</td>
<td>Lower one-third</td>
<td>2.5 × 1.0</td>
<td>Glomus tumor</td>
</tr>
<tr>
<td>Ito et alb</td>
<td>1988</td>
<td>51</td>
<td>M</td>
<td>Hemoptysis</td>
<td>Upper one-third</td>
<td>1.5 × 1.2</td>
<td>Glomus tumor</td>
</tr>
<tr>
<td>Present case</td>
<td>1989</td>
<td>47</td>
<td>F</td>
<td>Hemoptysis, cough</td>
<td>Above bifurcation</td>
<td>1.5 × 1.0</td>
<td>Oncocytic glomus tumor</td>
</tr>
</tbody>
</table>

Oncocytic Glomus Tumor of Trachea (Shin et al)
lamina and thin cytoplasmic microfilaments with dense bodies and dense attachment plaques on the cell membrane.

Ito et al reported a case of tracheal glomus tumor. There were some areas with an oncoticy appearance, and the tumor cells had eosinophilic and granular cytoplasm in which abundant mitochondria were electron microscopically present; but in most areas, the cytoplasm was clear, with ill-defined borders. In the present case, all of the tumor cells were characterized by strongly eosinophilic granular cytoplasm, resulting in well-defined cell outlines on light microscopy, and by numerous closely packed round mitochondria with prominent tubular cristae on electron microscopy. These pathologic findings had a strong resemblance to those of the case of Slater et al.7

The present report dealt with what we believe to be the first case of oncoticy glomus tumor arising in the trachea. The tumor cell was richly vascularized and bled easily, leading to hemoptysis. Careful handling during the biopsy procedure to prevent hemorrhage was necessary, and histologic confirmation was mandatory, with a local resection for the curative procedure.

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Mucoid Impaction of Upper Lobe Bronchi in the Absence of Proximal Bronchiectasis

William McD. Anderson, M.D., F.C.C.P.

Mucoid impaction is a complication of asthma and is frequently recurrent in patients with allergic bronchopulmonary aspergillosis (ABPA). This report describes a patient with asthma and recurrent bilateral well-circumscribed densities on chest roentgenogram in the absence of ABPA. Recognition of this clinical presentation may avoid the need for invasive diagnostic procedures or steroid therapy.

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ABPA = allergic bronchopulmonary aspergillosis

Transient bilateral densities associated with proximal bronchiectasis and mucoid impaction of bronchi are the roentgenographic hallmarks of allergic bronchopulmonary aspergillosis. This report illustrates the fact that recurrent bilateral perilunar densities due to mucoid impaction may occur in a patient with asthma in the absence of either bronchiectasis or hypersensitivity to Aspergillus species. Recognition of this entity and its response to appropriate therapy eliminates the need for invasive diagnostic procedures, including bronchoscopy and bronchography.

CASE REPORT

A 40-year-old woman was admitted to the hospital for evaluation of cough and fever associated with bilateral lung masses on chest roentgenogram. Two weeks prior to hospital admission she noted rhinorrhea, wheezing, and cough productive of small amounts of white sputum. Pleuritic chest pain, fever, and headache began two days prior to admission. She had a 25 pack-year smoking history and had worked only briefly. Gastric bypass surgery for obesity had been performed 15 years prior with reanastomosis later performed because of small bowel necrosis and infection. She continued to have intermittent symptoms of reflux esophagitis but without symptoms of aspiration. Two years prior, she was hospitalized for wheezing and bilateral perilunar densities on chest roentgenograms (Fig 1a). This illness responded to therapy with theophylline and penicillin (Fig 1b), and since that time, she has intermittently used theophylline.

She was obese and in moderate respiratory distress with a temperature of 38°C and absence of a paradoxic pulse. Percussion over the nasal sinuses did not reveal tenderness. The nasal mucosa was edematous. Tactile fremitus of the chest was normal with diffuse wheezing heard on auscultation. Egophony was not present. A chest roentgenogram showed bilateral, well-circumscribed perilunar densities (Fig 2a) as well as a left upper lobe nodule. Examination of sputum revealed mostly epithelial cells, and special stains and cultures for fungi and mycobacteria were negative. Her white blood cell count was 8,500 cells/μmm with 50 percent neutrophils, 40 percent lymphocytes, and 10 percent monocytes. Peripheral blood eosinophilia was not present and the serum IgE was 178 IU/ml with a negative serum precipitin titer for Aspergillus species. Breathing room air, arterial blood pH was 7.50, PaCO2 was 28 mm Hg, and