Pleomorphic Adenoma of the Trachea

A case of salivary gland type pleomorphic adenoma of the trachea in a 48-year-old woman is presented. Pleomorphic adenoma is extremely rare in the trachea. The behavior of this tumor in the trachea appears to be similar to its counterpart in other sites and distinctly different from the more frequently encountered epithelial tumors.
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INTRODUCTION

Tumors bearing the features of benign mixed tumors (pleomorphic adenoma) have been rarely recognized in the trachea, despite being the most common histologic form of salivary gland neoplasm (1-7). There are fewer than 40 cases in the literature (8). We report a case of primary mixed tumor of salivary gland-type, which was cytologically diagnosed, arising in the trachea.

CASE REPORT

A 48-year-old woman was admitted due to dyspnea on exertion and productive cough with wheezing for 3 months. Physical examination revealed decreased breathing sound in the left lung field. A chest x-ray on admission revealed suspicious narrowing of the distal tracheal lumen. A CT scan revealed a small polypoid intraluminal mass with homogeneous density in the left side of the distal trachea (Fig. 1). Bronchoscopy revealed a polypoid intraluminal tumor obstructing the tracheal lumen, just proximal to the carina (Fig. 2). A bronchoscopic needle aspiration was performed and it was interpreted as an adenoma (Fig. 3).

She underwent surgery and a well demarcated tumor, 1.5×1.2 cm in cross diameter, was resected by tracheal wedge resection procedure. The outer surface of the tumor was smooth, glistening, and well demarcated. The cut surface of the tumor was firm and cartilagenous, homogeneous gray and slightly mucoid. Microscopically, the surface of the tumor was covered by a mixture of respiratory epithelium and metaplastic squamous epithelium. The tumor was histologically characterized by a biphasic composition showing admixture in varying proportions of epithelial and stromal elements. The tumor showed the features classical of salivary gland-type mixed tumor with a glandular epithelial component and chondromyxoid stroma (Fig. 4). Some areas showed a paucity of glandular epithelial component and predominent solid

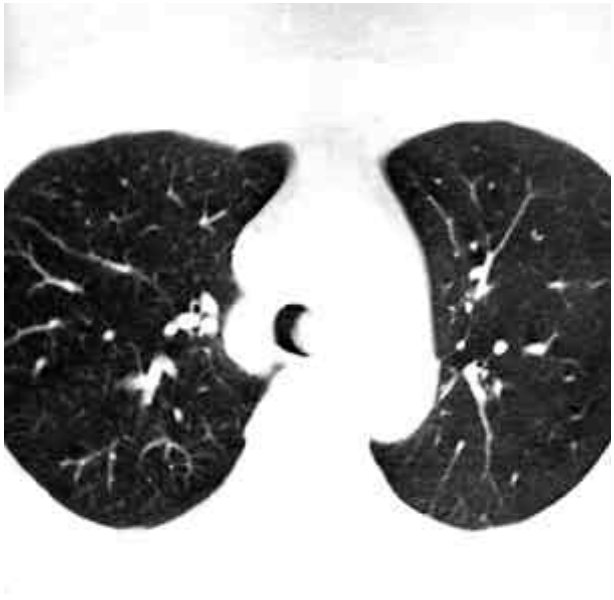

Fig. 1. CT scan obtained just above the tracheal carina shows a smooth marginated intratracheal mass arising from the left side wall.

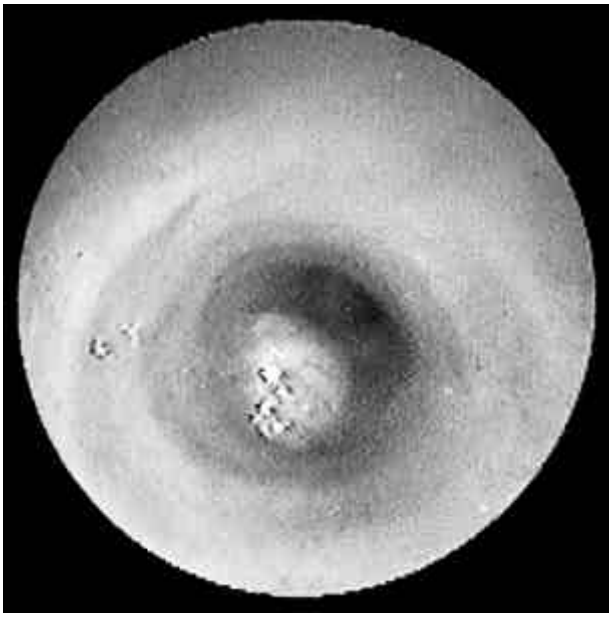

Fig. 2. Bronchoscopy shows an intraluminal polypoid mass with a smooth and glistening surface.

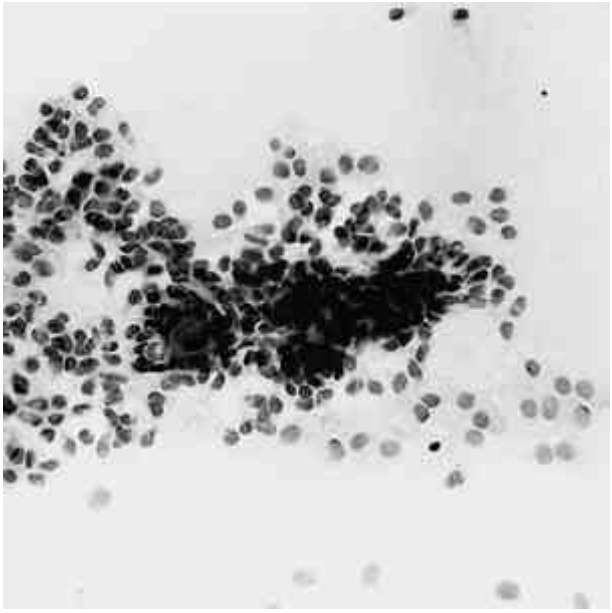

Fig. 3. Aspiration cytology reveals sheets of epithelial cells and individual myoepithelial cells in an amorphous myxoid background (H&E, ×200).

sheets or trabeculae of myoepithelial cells set against a prominent myxoid background. The stromal myxoid areas were strongly positive for alcian blue staining. Immunohistochemically, stromal and epithelial components were strongly positive for vimentin. The epithelial component was weakly positive for cytokeratin, S-100 protein, glial fibrillary acidic protein, and smooth muscle actin.

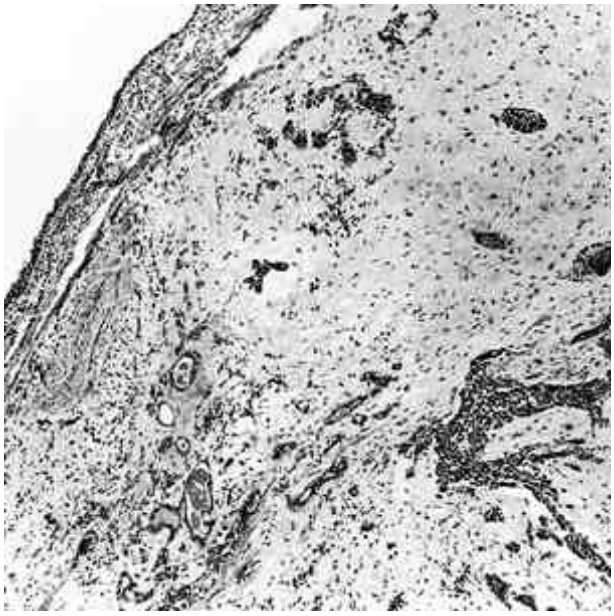

Fig. 4. Photomicrograph of the resected specimen shows intact respiratory or metaplastic squamous epithelium covering the surface of the tumor. The submucosal tumor was composed of epithelial or myoepithelial nests and abundant myxochondroid matrix (H&E, ×100).

DISCUSSION

The pathogenesis of pleomorphic adenoma of salivary gland type in the trachea remains a matter of controversy. Although the lesions traditionally are regarded as originating from the epithelium of submucosal glands (9, 10), their occurrence in peripheral or subpleural locations unrelated to a gland would seem to argue against this. Thus, the possibility exists that these neoplasms may originate from primitive stem cells, not necessarily restricted to the submucosal glands, bearing the capability to differentiate toward ductal structures, myoepithelium, and chondromyxoid matrix (1).

The average age of patients is 48 years with a range from 26 to 71 years (8). Tracheal and bronchial tumors usually present with intermittent episodes of productive cough and shortness of breath. Other symptoms are dyspnea on exertion, hemoptysis, wheezing, stridor, hoarseness, dysphagia, and recurrent respiratory tract infection. Peripheral tumors may be identified only on routine chest radiographs. The size of the tumor is variable. The tumors are generally sessile, whitish gray, firm, and polypoid. The cut surface is firm, gray, and slightly mucoid (1, 8).

Microscopically, tumors usually arise in the subepithelial tissues and are covered by a mixture of respiratory

and squamous epithelium. The glands have a tubular pattern, with a varying amount of eosinophilic material. Foci of squamous metaplasia have been identified in the tumor, and the epithelial elements are mixed with myxochondroid matrix. Foci of fibrosis are present. The stroma is rich in alcian blue-staining mucopolysaccharide (8). Tumor cells are usually positive for S-100 protein, cytokeratin, glial fibrillary acidic protein, muscle specific actin, and vimentin (1, 11).

The behavior of these tumors in the trachea appears to parallel those of their counterparts in salivary glands. Small, well-circumscribed lesions tend to behave in a benign fashion. Larger, more infiltrative or poorly circumscribed lesions will tend to recur and rarely metastasize (1).

The main differential diagnosis is adenoid cystic carcinoma which is a more aggressive tumor. Amorphous PAS-positive tubular material, absence of cartilagenous foci, perineural invasion, and the growing features of adenoid cystic carcinoma are distinct differential points from pleomorphic adenoma (8).

Mixed tumor of salivary gland type in the trachea can be diagnosed by aspiration cytology as could this case.

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