Unusual Intrapulmonary Tumor*
A Rare Cause of Bronchiectasis

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A 31-year-old black nurse was most recently admitted for assessment of recurrent hemoptysis and severe bronchiectasis. The patient had been well to age 15, when she began suffering recurrent respiratory tract infections and fevers. At ages 20 and 22 years, partial right pneumonectomies were performed for recurring hemoptysis.

Chest radiographs demonstrated extensive bronchiectasis in the right lung associated with fibrosis and volume loss. Punctate calcifications were noted in the right suprahilar and paratracheal regions where there was severe fibrotic and architectural distortion. An air space opacity was identified in the lingula with possible underlying bronchiectasis. The remainder of the left lung appeared normal (Fig 1). Fiberoptic bronchoscopy revealed edematous granular mucosa with copious purulent secretions in all of the visualized right bronchial tree. No abnormality was identified in the left system.

Computed tomography (CT) of the chest confirmed extensive destructive and cystic disease in the right lung and bronchiectasis limited to the lingula in the left lung (Fig 2). A mass was demonstrated in the right upper hemithorax; it did not enhance after intravenous contrast was injected, but contained punctate calcifications and scattered areas of decreased CT density. The mass extended into the superior mediastinum surrounding the right brachiocephalic vein and engulfing the lower superior vena cava. A fine needle percutaneous aspiration biopsy was performed, and cytologic study revealed respiratory epithelium, squamous epithelium and mesothelium. No malignant cells were identified.

Right pneumonectomy was carried out. Surgery was complicated by the presence of dense fibrous and hyperemic scar tissue binding the entire lung to the chest wall. Despite maximal surgical attempts to stop hemorrhage, the patient continued to bleed and expired 48 hours following surgery.

Histologic examination of the resected specimen showed well differentiated tissues representing all three germinal layers (Fig 3). Organoid arrangements of skin appendages, brain, bone, fat, muscle and

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gastrointestinal tissue were present. Pancreatic islet and acinar tissue was prominent. There was direct continuity of tumor with the lung parenchyma, and its mediastinal aspect was covered by visceral pleura. The remaining lung was grossly bronchiectatic.

**Diagnosis: Benign intrapulmonary teratoma and ipsilateral cystic bronchiectasis**

Intrathoracic teratomas almost always occur in the mediastinum, and only very rarely arise within the lung. By 1978, Holt et al found 13 histologically confirmed lung teratomata in the world literature. Since then, three have been added to the English, and one to the German literature. Seven tumors have been located in the left upper lobe, three in the right upper lobe, and six confined to the middle and lower lobes; in one, the exact intrapulmonary site was not listed. Eleven tumors have been malignant and six benign. In our patient, the tumor was benign and situated in the right upper lobe. Though extension into the mediastinum was demonstrated, histologic evidence of a primarily intrapulmonary location was convincing. Sections showed pancreatic tissue in direct continuity with bronchioles and cavities; further evidence of this included the presence of a tuft of hair protruding from the upper lobe bronchus on the resected specimen. Reported symptoms associated with pulmonary teratoma have been prolonged and have included cough, hemoptysis, chest pain and trichoptysis. Plain roentgenographic features are of a calcified mass, which may show peripheral translucent areas. In our patient, though calcification was seen, even in retrospect the mass was not discernible on the chest radiographs due to destruction of the contiguous lung. CT appearances were more helpful, indicating a mass of complex structure containing punctate calcification and areas of high local fat content. CT also demonstrated the degree of mediastinal invasion and relationship of the tumor to vascular structures.

One previous report has described bronchiectasis in association with intrapulmonary teratoma. The authors postulated that long term drainage of infected material produced by local tumor compression was the cause.

Because of the prominence of pancreatic glandular tissue in our patient's tumor, we suggest that bronchiectasis resulted from digestive enzyme activity on the draining bronchial tree. This hypothesis is supported by the absence of other causes for such extensive bronchiectatic change. Results of repeated cultures for acid-fast bacilli, and tuberculin skin tests were documented to be negative during the six years the patient attended this hospital. Though positive skin tests to *Histoplasma capsulatum* were demonstrated, the patient had previously had a full course of therapy with amphotericin B, and bronchiectasis progressed despite repeated negative Histoplasma cultures and stains. The distribution of bronchiectasis was compatible with such a mechanism, showing massive right lung involvement with sparing of the left lung, apart from the lingula.

In retrospect, the tumor was not convincingly appreciable on the plain chest radiograph. The lesion was clearly visible on CT and, had the entity been considered, a diagnosis was possible from percutaneous biopsy histology results.

Pulmonary teratoma is a rare tumor, and we report a case where associated bronchiectasis was a very prominent feature. A relationship between the two conditions in this patient is postulated and the literature reviewed. Awareness of the tumor in the lung and its possible autodigestive properties would have altered management and possibly prevented further bronchial destruction. If the mass contains cartilage and a high fat content, the CT appearances can be expected to be characteristic. We will consider the entity if a similar clinico-radiologic situation is again seen.

**References**

5. Eckert VM, Gerassimidis T. Intrapulmonales teratmon. Fallbericht und Literaturubersicht Fortschr Med 1979; 97:1051-54
6. Ruland L. Malignant teratoblastoma of the lung. Thoraxchirurgie 1956; 4119-21