Case Report

Primary Pulmonary Teratoma: Report of a Case and the Proposition of "Bronchotrichosis" as a New Term

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Primary pulmonary teratoma is a very rare disease. Most follow a benign course and are incidental findings during routine chest X-rays. Hair found in sputum or in bronchus detected during bronchoscopy is also a rare condition and is usually caused by mediastinal teratoma. This case report is of a 36-year-old man who presented with halitosis. A fiber-optic bronchoscopy revealed coarse hair originated from the right upper lobe. The patient was successfully treated by right upper lobectomy, and pathology confirmed primary pulmonary teratoma. We recommend that "bronchotrichosis" could be used as a new term for such a sign. (Ann Thorac Cardiovasc Surg 2009; 15: 247–249)

Key words: primary pulmonary teratoma, lobectomy

Introduction

Various intrapulmonary benign tumors can produce compressive symptoms and signs in patients bearing such tumors.[1)] However, the invasion of pulmonary structures by benign tumors is rarely seen.[1)] The resection of a discovered pulmonary benign tumor is advisable when feasible in terms of a patient's functional status and complete resectability of the lesion.

Examples of primary intrapulmonary teratoma have been reported only with extraordinary rarity.[2,3)] Primary pulmonary teratomas that lack cytologically malignant components are so uncommon as to be anecdotal.[4)]

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Case

A 36-year-old male was presented to a chest disease clinic with complaints of halitosis. A chest radiograph disclosed a right upper lobe mass, and a chest computed tomography revealed a 6-cm mass at the right upper lobe with mediastinal extension (Fig. 1). A fiber-optic bronchoscopy revealed condensed and coarsened hair. Only brown hair could be extracted during bronchoscopic biopsy (Fig. 2), and no tissue biopsy was possible. A transthoracic needle aspiration of the lesion was evaluated to be nondiagnostic. The material consisted of lymphocytes and histiocytes with no diagnostic suggestion. The patient was thought to have teratoma, and an anterior thoracotomy was performed for resectional surgery. On exploration, a right upper lobe mass with anterior mediastinal adhesions was anticipated. The lesion was freed from superior vena cava, and a lobectomy was achieved. Hair was seen after the bronchus was cut. Pathological examination revealed a mature teratoma containing derivatives from ectoderm, endoderm, and mesoderm. Bronchus epithelium with sebaceous glands and hair follicles were seen microscopically (Fig. 3). Since there was subepithelial sebaceous glands and hair follicles with clear pulmonary

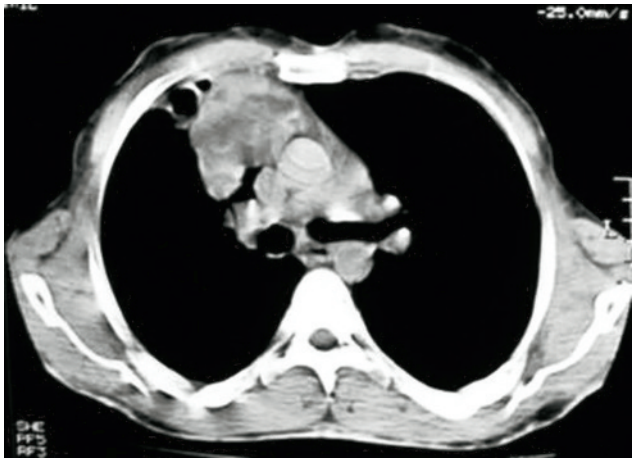

Fig. 1. Chest computed tomography scan.
A pulmonary mass adjacent to mediastinal structures is seen.

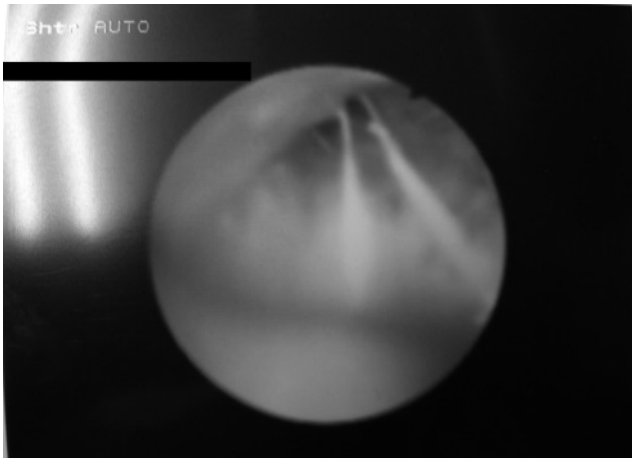

Fig. 2. Bronchoscopic view of hair emerging from the right upper lobe bronchus.

parenchyma areas adjacent to the mediastinal pleura, the diagnosis was primary pulmonary teratoma. The postoperative course was uneventful, and the patient has been doing well for 1.5 years.

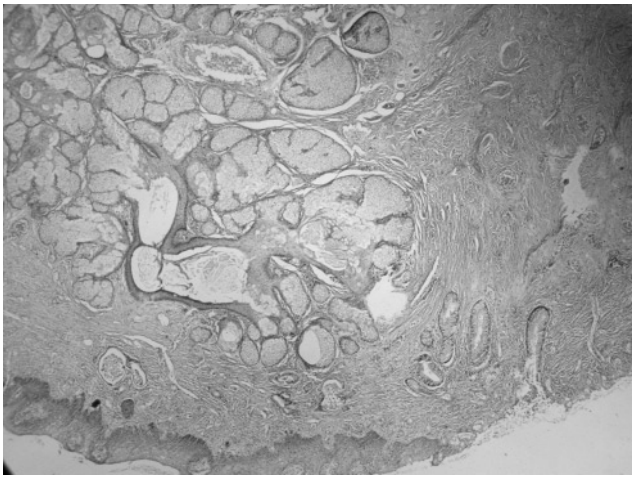

Fig. 3. A view of primary pulmonary (mature) teratoma showing subepithelial hair follicles and sabeceous glands and ductus. (hemotoxylin-eosin; original magnification: ×40)

Discussion

Teratomas are tumors consisting of tissues derived from more than one germ cell line. They may be mature or immature. Almost all teratoid tumors in the lungs and pleura represent altered metastases of gonadal germ cell tumors, in which malignant histological components were formerly present, but they can be completely cured by chemotherapy.[4] Another small group of cases comprises primary teratoid tumors of the lung, including seminoma, embryonal carcinoma, yolk sac carcinoma, and choriocarcinoma. Although mediastinal teratomas are far from being a rarity, primary pulmonary teratomas that possess no malignant features are extremely rare. A total of 67 cases have been reported in the literature from 1939 to 2007, including 35 from Japan and 7 from Korea, and the rest are in the English literature.[2–9] Tandon and colleagues[3] reported on a patient with primary pulmonary teratoma presenting as pyothorax. In our patient, a postoperative macroscopic examination of the specimen unveiled an oily, yellowish infected material, including hair, that was noticed inside the lesion. The lesion was seen to be associated with the upper lobe bronchus.

Direct bronchial communication is a distinguishing feature of intrapulmonary teratomas and has been reported in most of the articles.[2–9]A cavity with peripheral translucency is a feature distinguishing intrapulmonary teratomas from mediastinal teratomas.[8,9] This feature indicates air within the cavity arising from bronchial communication.

CT scans demonstrate discrete areas of soft tissue, high local fat content, punctate calcification, or a combination of these areas and is extremely valuable to detect a ruptured teratoma.[7] In ruptured teratoma, the internal density becomes heterogeneous, the tumor margin becomes irregular, and the fat component shows a bursting configuration.[9] The radiological findings in our case are in complete concordance with these features. And in our case, a separate cavity rather than mediastinal tumoral involvement was noted. Intrapulmonary terato-

mas are predominantly located in the upper lobe (67%), mostly in the anterior segment, as we found in our patient. Most have occurred in the first or second decade of life, the ages ranging from 10 to 68 years.[9] Moreover, our case had no history of pleural infection. Nevertheless, halitosis could be due to the infectious teratomatous tissue, and it disappeared after surgical resection.

Mesodermal, ectodermal, and endodermal elements are seen in teratomas in varying proportions. Most pulmonary teratomas are composed of mature, often cystic, somatic tissue. In our patient, tissues that are remnants of embryonic derivatives of hair, stomach, and mammary gland were seen. Hairy content of the lesion is attributable to the presence of derivatives from the ectodermal line.[2] Since primary pulmonary teratoma could produce massive intrabronchial hair, we would like to propose "bronchotrichosis" (*trichosis* is Latin for hair) as a new term for such a sign. As shown in Fig. 2, hair follicles were located just under the bronchial epithelium. For this reason it must be discriminated from hair expectoration, which could happen because of a pulmonary invasion of mediastinal teratoma. In our case, hair production is just beneath the bronchial epithelium. It is fair to recommend such a term, since subepithelial hair follicles are seen microscopically.

In conclusion, the discovery of primary intrapulmonary teratomas is extremely rare. "Bronchotrichosis" is the only clinicopathological feature that can be diagnostic. It can be cured by anatomical lung resection.

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