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. However, the invasion of pulmonary structures by benign tumors is rarely seen. 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. Primary pulmonary teratomas that lack cytologically malignant components are so uncommon as to be anecdotal.

**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 trans-thoracic 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...
parenchyma areas adjacent to the mediastinal pleura, the
diagnosis was primary pulmonary teratoma. The postop-
erative course was uneventful, and the patient has been
doing well for 1.5 years.

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 com-
prises 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
colleagues3) reported on a patient with primary pulmo-
nary 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 combi-
nation 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 burst-
ing 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-

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.

Fig. 3. A view of primary pulmonary (mature) teratoma showing
subepithelial hair follicles and sebaceous glands and ductus.
(hemotoxylin-eosin; original magnification: ×40)
Primary Pulmonary Teratoma: Report of a Case and the Proposition of “Bronchotrichosis” as a New Term

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. 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. 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.

References