

Intrapulmonary Teratoma: A Report of Three Cases

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Abstract

Three cases of intrapulmonary teratoma are described. A specific symptom of trichoptysis occurred in two patients. The other presented with only recurrent hemoptysis. All were treated by lobectomy of the affected lung. Pathologic examination in each specimen showed that the tumor had a cystic portion which was connected to the bronchial system and contained sebaceous material and pieces of hair. The solid portion consisted of tissues representing all three germ cell layers. The pathologic findings correlated well with the clinical symptoms and radiologic findings in the patients.

Teratomas are true neoplasms made up of tissues from one or more of the three germ cell layers. They are usually found in the ovary, testis and mediastinum. Those arising in the lung are very rare. Here we describe 3 cases of intrapulmonary teratoma with clinicopathologic correlation.

CASE REPORTS

Case 1

A 45-year-old Thai male was admitted to the Central Chest Hospital due to hemoptysis. For

seven years, the patient had not only recurrent symptoms of hemoptysis but also chronic cough and sometimes he coughed up pieces of hair. One day before, he had expectorated about 100 ml of fresh blood. On physical examination, no abnormal clinical findings or obvious source of bleeding were detected. Chest X-ray revealed haziness of the left upper lung field with sharp convex border, consistent with an intrapulmonary mass (Fig. 1). Fiberoptic bronchoscopy revealed narrowing of the segmental bronchus to the left upper lobe of the lung and oozing of blood through the bronchial

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lumen. Transbronchial lung biopsy was performed and demonstrated pieces of hair which could be examined macroscopically and inflamed bronchial mucosa examined under microscopy. Thoracotomy with left upper lobe lobectomy was performed and the mass was removed completely. The patient recovered well after surgery. There was no evidence of recurrence or metastasis on follow-up.

Grossly the specimen of the left upper lobe of the lung, measuring 21x11x4 cm and weighing 220 g, revealed an intrapulmonary cystic mass with connection to the segmental bronchus. The cyst, measuring 5 cm in greatest dimension, contained pieces of hair, sebaceous material, necrotic tissue and a solid portion of fibrofatty tissue (Fig. 2A). The cystic wall was fibrotic and rather smooth. The surrounding lung tissue revealed bronchiectasis and congestion.

Microscopically the cystic wall was lined by keratinizing stratified squamous epithelium. Some parts of the epithelium were intact but some were disrupted with granulation tissue formation. The solid portion consisted of various differentiated types of tissue: skin, skin appendages including hair, fibroadipose tissue and respiratory epithelium (Fig. 2B). The pathologic findings were typical of intrapulmonary mature cystic teratoma.

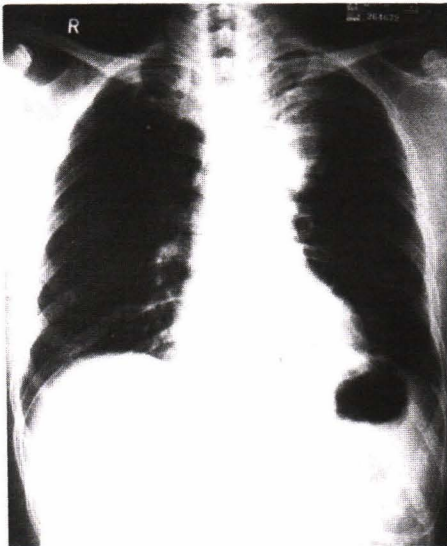


Fig. 1. Chest X-ray from case 1 showing a mass shadow confined to the left upper lobe of lung.

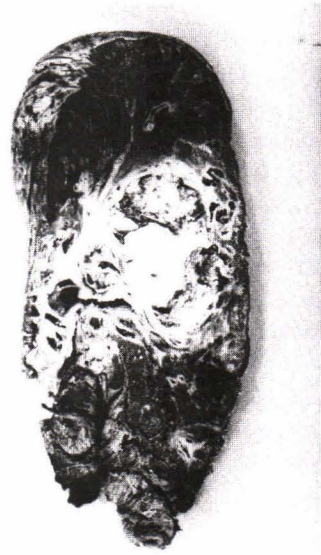


Fig. 2A. Surgical specimen from case 1 showing connection of the segmental bronchus to the intrapulmonary cyst containing pieces of hair, sebaceous material, necrotic tissue and a solid portion of fibrofatty tissue.

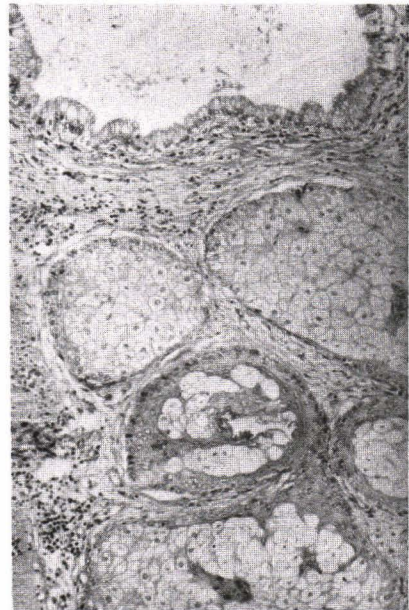


Fig. 2B. Microscopically, the tumor from case 1 showing skin appendage differentiation admixed with respiratory epithelium with goblet cell metaplasia.

Case 2

An 18-year-old Thai female went to the hospital due to problems of chronic cough, recurrent hemoptysis and expectoration of hair for 12 months. She was previously diagnosed as having pulmonary tuberculosis and was completely treated with a short-course-regimen of antituberculous drugs at a local hospital with no clinical improvement. On physical examination, no abnormal clinical findings or obvious source of bleeding were noted. Chest X-ray revealed a patchy shadow with air bronchogram and a small cavity at the right middle lung field. Computed tomography of the chest revealed consolidation with air bronchogram and small cavities at the right middle lobe of the lung with continuation to an inhomogeneous mass at the medial side. The mass, measuring 5 cm in greatest dimension, showed various soft tissue densities including fat density and was confined to the right hemithorax (Fig. 3). Fiberoptic bronchoscopy failed to demonstrate any endobronchial lesions. Thoracotomy was performed and revealed that the right middle lobe of the lung was fixed to the mass which extended to the mediastinum. Right middle lobe lobectomy with removal of the mass was done. The patient recovered well after surgery and showed no evidence of recurrence or metastasis on follow-up.

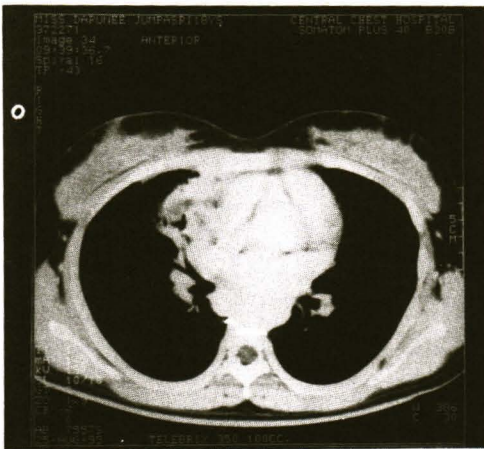


Fig. 3. Computed tomography of the chest from case 2 showing consolidation with air bronchogram and small cavities of the right middle lobe of the lung with continuation to an inhomogeneous mass at the medial side.



Fig. 4. Surgical specimen of the lung from case 2 showing a pale solid region of lipid pneumonia surrounding the intrapulmonary cystic portion of the tumor containing sebaceous material with continuation to the solid portion extending out of the lung.

Grossly the specimen consisted of the right middle lobe of the lung, attached to a 5x3.5x3.5 cm well-circumscribed mass, covered with continuing visceral pleura at the medial part. Cut surfaces of the lung revealed yellowish gray firm airless lung tissue surrounding an intrapulmonary cyst measuring 1.3 cm in diameter with connection to the segmental bronchus. The cystic cavity was filled with sebaceous material and tiny pieces of hair. The cystic wall was fibrotic and continued to intrapulmonary solid gray-white portion of the tumor then continued to a larger solid portion which extended out of the lung. Cut surfaces of this portion revealed solid fibrofatty tissue with gray-brown tissue and pieces of hair (Fig. 4).

Microscopically the intrapulmonary cystic portion of the tumor was lined by keratinizing stratified squamous epithelium admixed with ciliated pseudostratified columnar epithelium and granulation tissue. Some pieces of hair arose from the cystic lining. The continuing intrapulmonary solid gray-white portion consisted of mostly sclerotic vascular structure and a little adipose tissue. The extrapulmonary solid portion consisted of various differentiated types of tissue: skin and skin appendages, adipose tissue, bone, bone marrow, respiratory epithelium, pancreatic tissue and thymic tissue. The lung parenchyma surrounding the cystic portion of the tumor revealed lipid pneumonia. The

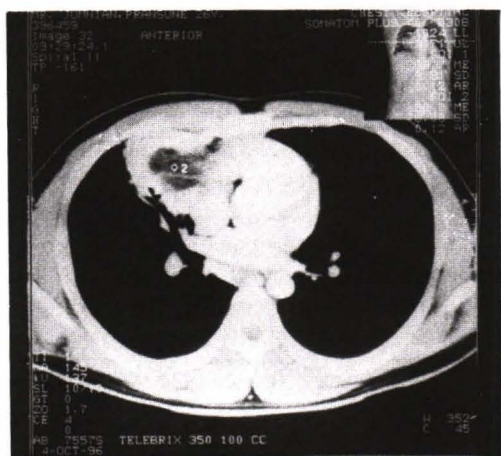


Fig. 5. Computed tomography of the chest from case 3 showing patchy infiltration with air bronchogram and some cavities at the right middle lobe of the lung with continuation to an inhomogeneous mass at the medial side.

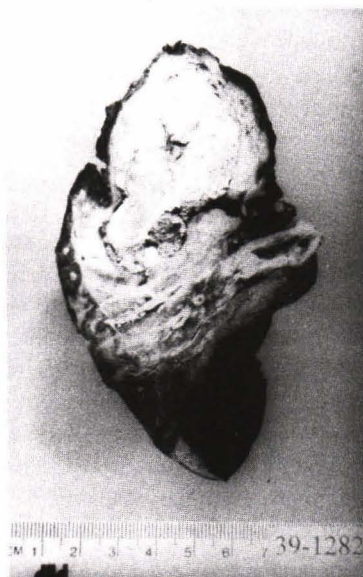


Fig. 6. Surgical specimen of the lung from case 3 showing an intrapulmonary cyst packed with sebaceous material, pieces of hair and a solid portion of fibrofatty tissue.

pathologic findings were consistent with a mature cystic teratoma arising from the bronchus with solid portion extending to the mediastinum.

Case 3

A 26-year-old Thai male went to the hospital due to recurrent hemoptysis for 2 years. Physical examination revealed no abnormal clinical finding. Chest X-ray revealed a mass shadow of the right middle lung field. Computed tomography of the chest revealed patchy infiltration with air bronchogram and some cavities at the right middle lobe of the lung which continued to an inhomogeneous mass with various soft tissue densities including fat density, measuring 6 cm in greatest dimension at the medial side (Fig. 5). Fiberoptic bronchoscopy revealed inflamed bronchial mucosa of the right middle lobe of the lung. Thoracotomy with right middle lobe lobectomy was performed. The patient recovered well and had no recurrent or metastasis on follow-up.

Grossly the specimen of the right middle lobe of the lung, measuring 12x10x4.5 cm and weighing 210 g, showed a well-circumscribed intra-

parenchymal mass. The mass, measuring 6 cm in greatest dimension, had a protruding portion with complete visceral pleura covering. Cut surfaces of the mass revealed a large cystic cavity containing fatty tissue with heterogeneous grey-brown tissue and packed with sebaceous material and pieces of hair (Fig. 6). The cystic cavity was connected to the right middle lobe bronchus which also contained sebaceous material in the lumen.

Microscopically the cystic wall was lined by keratinizing stratified squamous epithelium and granulation tissue. The solid portion revealed differentiated types of tissue: skin, skin appendages, fibroadipose tissue, cartilaginous tissue, respiratory epithelium, seromucinous gland and thymic tissue. The pathologic findings were typical of intrapulmonary mature cystic teratoma.

DISCUSSION

Intrathoracic teratomas arising in the mediastinum are not uncommon but those arising in the lung are rare neoplasms⁽¹⁾. Morgan et al in 1992 reported a case and reviewed the 30 previously reported cases of intrapulmonary tera-

tomas⁽²⁾. Since then, there have been 2 additional reported cases⁽³⁾. In Thailand, there was only one published case in 1975⁽⁴⁾. Most of the patients had symptoms of chest pain, hemoptysis and cough. Only a few cases complained of trichoptysis or expectoration of hair⁽²⁾. In our patients, the ulcerated epithelial linings of the cystic walls with granulation tissue formation were the main sources of bleeding. Pieces of hair, squame cells and sebaceous material, the ectodermal components of the well-differentiated tumors, were collected in the cystic cavities which had connection to the bronchial lumens. When the patients coughed, they coughed up not only an amount of fresh blood but also some pieces of hair and necrotic tissue. These described how hemoptysis and trichoptysis occurred. In case 2, the patient developed not only a symptom of trichoptysis but also aspiration of hair and squame cells and resulted in lipid pneumonia of the lung surrounding the cyst.

Bronchoscopy is a useful diagnostic procedure for tumors that had connection to the bronchial system which were found in less than half of the patients⁽²⁾. Demonstration of hair in the bronchial tree, equivalent to the rare symptom of trichoptysis, led to the preoperative diagnosis of intrapulmonary teratoma⁽⁵⁾. Bronchoscopy not only revealed pieces of hair, the important evidence for diagnosis of teratoma in case 1 but also revealed the site of the bleeding tumor which was useful for the following management.

Computed tomography of the chest was also a useful investigation but has been reported in only a few cases^(2,6,7). The findings included

homogeneous or heterogeneous masses depending on their tissue components. The heterogeneous masses showed foci of calcification and various soft tissue densities including fat density. All masses occupied the hemithorax and showed some areas adjacent or extending to the mediastinum which could lead to misdiagnosis as mediastinal tumors. In case 2, computed tomography revealed consolidation due to lipid pneumonia of the lung tissue surrounding the cystic portion of the tumor. The small intrapulmonary portion of the teratoma was not clearly defined in comparison with the larger extrapulmonary portion which demonstrated more details. We considered our second case as an intrapulmonary teratoma with mediastinal extension instead of mediastinal teratoma with lung invasion because the latter should be an immature teratoma, teratocarcinoma or teratoma with malignant transformation which had ability of tissue invasion. Pathologic examination revealed that all components of the tumor were benign and mature. The intrapulmonary cystic portion with bronchial connection which consisted of mature ectodermal component resembling skin and skin appendages and the intrapulmonary solid portion which consisted of mature components of mesodermal differentiation could not be the result of any tumor invasion but arising from the lung itself.

Surgery was an appropriate treatment in our cases because the bleeding tumors could be removed completely and the lesions could be examined histologically. The pathologic findings of the tumors correlated well with the clinical symptoms and signs of these patients.

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เทอร์าโทมาในปอด: รายงานผู้ป่วย 3 ราย

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รายงานผู้ป่วย 3 รายที่ได้รับการวินิจฉัยว่าเป็นเทอร์าโทมาในปอด พบอาการไอเป็นชนซึ่งเป็นอาการเฉพาะสำหรับโรคนี้ในผู้ป่วย 2 ราย อีกหนึ่งรายมีเพียงอาการไอเป็นเลือด ผู้ป่วยทั้ง 3 รายได้รับการผ่าตัดเอาปอดกลับที่เป็นโรคออก จากการตรวจทางพยาธิวิทยา พบโพรงของเนื้องอกติดต่อกับหลอดเลือด และถูกอัดแน่นด้วยคราบไขมัน และเส้นขน ส่วนของเนื้องอกที่เป็นก้อน ประกอบด้วยเนื้อเยื่อที่เจริญจาก germ cell ทั้ง 3 ชั้น ลักษณะสิ่งตรวจพบทางพยาธิวิทยา อธิบายอาการของผู้ป่วย และภาพรังสีของทรวงอกได้เป็นอย่างดี

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