Intrapulmonary Cystic Benign Teratoma: A Case Report and Review of the Literature

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We describe a 41-year-old woman with a short history of retrosternal chest pain and non-productive cough due to a benign intrapulmonary teratoma originating from the left upper lobe. The clinical, CT features of this rare tumor are presented and the relevant literature is discussed. (Ann Thorac Cardiovasc Surg 2004; 10: 290–2)

Key words: teratoma, intrapulmonary tumor, dermoid cyst

Introduction

Intrathoracic teratomas almost always occur in the mediastinum and rarely arise within the lung. Until 1996 approximately 30 cases of intrapulmonary teratoma (IPT) have been reported in the English literature.1 The majority of these tumors are benign although malignant tumors have been described, too.2 Typically, cystic teratomas are found in the body’s midline. Common locations include the gonads, the mediastinum and the pineal region.

Reported symptoms associated with IPT include fever, cough, hemoptysis, chest pain and hair expectoration (trichoptysis).3 In rare instances, rupture into adjacent structures like pleural space, tracheobronchial tree or pericardium may occur.3 In addition, bronchiectasis and obstructive pneumonia may develop.4

We describe the case of a 41-year-old woman with a benign cystic teratoma originating from the left upper lobe, review the clinical and radiographic features and discuss the pertinent literature.

Case Report

A 41-year-old woman presented with an eight week history of thoracic pain and non-productive cough. She denied weight loss, fever, night sweats and hemoptysis. Her general practitioner referred her to a radiologist for further diagnostic work-up.

Chest x-ray demonstrated a perihilar partially air-filled cystic tumor in the left lung measuring approximately 5×4 × 4 cms (Fig. 1). A cavity was visible. The margins were sharp and no further pathology was observed. A spiral CT showed a cystic, rather homogenous tumor with a small spherical fatty component at the posterior lower circumference. This was considered a fat capsule. No calcification was noted. No connection with the tracheobronchial tree was identified but with the ventrolateral pleura (Fig. 2). The tumor was thought to be either a cystic teratoma or a pericardial cyst. Routine laboratory testing was entirely normal.

The patient was admitted to our hospital for further treatment. The tumor was approached via a left anterolateral thoracotomy (submammary incision) through the fifth intercostal space. A firm pedunculated tumor of approximately 6×4×4 cms was identified in the left upper lobe. There was no connection to any mediastinal structures or to the pleura. The tumor was excised and opened on the back table. It contained yellowish sebaceous material as well as some hair and fat (Fig. 3). The specimen was sent to our pathologists who confirmed a cystic lesion lined by squamous epitheliums with sebaceous and sweat glands without malignancy.

The patient’s postoperative course was uneventful and she was discharged home on postoperative day five.

Discussion

Intrathoracic teratomas almost always occur in the medi-
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Fig. 1. Chest x-ray with left-sided perihilar cystic tumor. Note the partial filling with air and the cavitation.

Fig. 2. CT scan displays a rather homogenous thin-walled tumor with a small fatty capsule at the posterior lower circumference. No calcification is visible. No connection with the tracheobronchial tree was identified.

Fig. 3. Intraoperative photograph of resected specimen demonstrating sebaceous material and hair.

astinum and only very rarely arise within the lung. Most authors have stated that the majority of tumors are located in the left lung.23) This was contradicted by Asano and coworkers.4) However, there is a predilection for the upper lobes on both sides. Due to this predilection it is thought that IPTs develop in relation to the thymus as derivatives of the third pharyngeal pouch. The presence of IPT may be the result of displacement or separation of the thymus during early embryogenesis.6)

According to Asano et al.4) 65 cases have been reported in the literature from 1839 until 1996, including 35 cases in Japan. IPT occur in adults as well as in children, the age ranging from 10 months to 68 years.8) The majority of patients are diagnosed in their first or second decade of life. The prevalence of IPT in women and men is similar.

Most symptoms like chest pain, fever, cough, hemoptysis, bronchiectasis, pneumonia, weight loss are non-specific and make the preoperative diagnosis extremely difficult. The bronchoscopic evidence of hair in the bronchial tree or the clinical equivalent of trichoptysis is considered a suggestive symptom. However, trichoptysis has been reported in seven patients only1,4) indicating the low incidence of this important symptom.

Typical x-ray findings include a lobulated opacity within the affected pulmonary lobe, calcification within the lesion,7) cavitation8) or peripheral translucent areas.9) The margin is usually smooth with a thin wall. In addition, consolidation of the lung may be noted. In our patient such a cavity with a peripheral translucent area was identified. It indicates air within the cavity originating from a communication of the teratoma and the bronchial system. Holt et al.10) used this feature to radiologically distinguish IPT from mediastinal teratoma. The presence of an air-filled cavity has several differential diagnoses like fungal mass, hydatid cyst or lung abscess.8) CT exams can facilitate further clarification by demonstrating
punctate calcification, discrete areas of soft tissue and areas of high local fat content. Our patient had a fatty capsule around the tumor (Fig. 2). CT is extremely valuable for the differentiation between a ruptured and an unruptured teratoma. Ninety percent of unruptured teratomas exhibit homogenous internal density. If rupture occurs, the internal density becomes heterogenous, the tumor margins change from smooth to irregular and the fat component from spherical to a bursting configuration. Furthermore, CT can demonstrate the degree of mediastinal invasion and the relationship to vascular structures.

IPT is characterized by different derivatives of endodermal, mesodermal and ectodermal origin. Due to this variety of tissue contained within IPTs, such tumors can produce proteolytic or digestive enzymes making them prone to rupture.

Owing to its potential for rupture surgical resection of any IPT is recommended. The other reason for resection is the high percentage (approximately 30%) of malignant teratomas. This definition has often been based on the presence of immature tissue within the teratoma rather than on presence of metastases or infiltration. This may be a confounding factor for the excellent results after surgical resection of some IPTs thought to be malignant. Interestingly, malignant teratoma was described more frequently in women (12 patients) than in men (5 patients).

In conclusion, any IPT should be resected due to its potential for malignancy and rupture. The diagnosis is obvious if hair is expectorated or detected in the bronchial tree. Since these are rare findings, the diagnosis has to rely on x-ray studies which typically demonstrate calcification, cavitation and peripheral translucent areas. CT examinations are helpful to distinguish IPTs from other air-filled intrapulmonary masses.

References