

## 폐동맥을 침범한 파열된 성숙 낭포성 기형종

— 1예 보고 —

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### Ruptured Mature Cystic Teratoma Involving Pulmonary Artery

— A case report —

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We report a case of mature cystic teratoma of the anterior mediastinum that ruptured into the pulmonary artery requiring an emergent surgical treatment. A 39-year-old woman presented an episode of massive hemoptysis and treated with bronchial artery embolization (BAE). On the 10th day after BAE, however, she developed sudden massive hemoptysis and had a deteriorated mental status. For a definitive treatment, she underwent the left pneumonectomy and the tumor resection in the anterior mediastinum. On histologic examination, the tumor disclosed cystic structures composed of mature squamous epithelium, pilosebaceous glands, mature fat tissue, cartilage and bone tissue. Also, there was intrapulmonary hemorrhage due to left pulmonary artery invasion of the tumor.

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**Key words:** 1. Teratoma  
2. Hemoptysis  
3. Lung

Benign teratomas of the mediastinum (mature cystic teratoma or "dermoid" tumor) are rare and account for only 3 to 12% of mediastinal tumors[1]. Although these tumors have been described in patients with ages ranging from 7 months to 65 years, most occur in young adults, with an approximately equal distribution in males and females[1,2]. Approximately 95% of benign teratomas arise in the anterior mediastinum; the remainder arise in the posterior mediastinum[1,2]. On rare occasions, cystic teratomas rupture into adjacent structures such as the pleural space, pericardium,

lung or tracheobronchial tree[3]. We experienced a patient with ruptured mature cystic teratoma involving pulmonary artery requiring an urgent surgical treatment and we report this case with a brief review of the literature.

### CASE REPORT

A 39-year-old woman was admitted to the local hospital with hemoptysis. During the admission, she developed one episode of massive hemoptysis. Her mental status was deterio-

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본 논문의 저작권 및 전자매체의 지적소유권은 대한흉부외과학회에 있다.



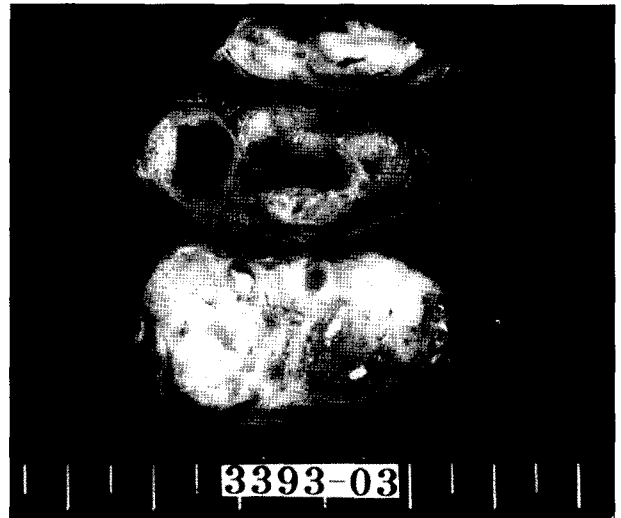
**Fig. 1.** Chest radiography shows diffuse increased density of the left lung and multiple air-space consolidations in right lower lobe.

rated and she was immediately intubated. Twenty years ago, she had a history of pulmonary tuberculosis and was successfully treated with a standard regimen for 9 months. The patient was transferred to Chonnam National University Hospital.

On arrival, she was alert with endotracheal intubation. A physical examination showed a temperature of 36.5°C, a blood pressure of 140/70 mm Hg, a pulse rate of 110 /min. On auscultation of the lungs, there was decreased breath sound in the left lung field. An abdominal examination revealed no remarkable findings. The values obtained during the initial laboratory examination were as follows: WBC count 8,900 cells/mm<sup>3</sup>, hemoglobin 7.9 g/dL, hematocrit 25.4%, platelet count 337,000 cells/mm<sup>3</sup>. The liver function tests, prothrombin and partial thromboplastin times were all within normal limits. The arterial blood gases on 5 L/min oxygen were pH 7.158, PaCO<sub>2</sub> 63.0 mm Hg, PaO<sub>2</sub> 71.5 mm Hg. A chest radiography showed diffuse increased haziness density of the left lung and multiple air-space consolidations in the right lower lobe (Fig. 1). Computerized tomography (CT) scanning demonstrated a 4 cm heterogeneously enhancing mass with dense calcifications in the left mediastinum



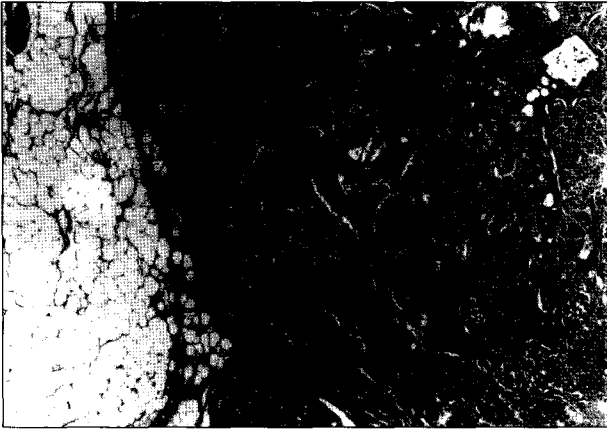
**Fig. 2.** Computerized tomography (CT) scanning demonstrates a 4 cm heterogeneously enhancing mass with dense calcification in the left mediastinum and multicystic changes with air-fluid levels in the left lung. Note the volume loss of left hemithorax with mediastinal shifting.



**Fig. 3.** The specimen shows well demarcated 7×5×2.3 cm sized mass composed of fat, fibrous tissue, cystic portion, and calcification.

and multicystic changes with air-fluid levels in the left lung (Fig. 2).

She was initially treated with bronchial artery embolization (BAE) and her symptoms were improved. On the 10th day after BAE, however, she developed massive hemoptysis and confused mental status. For a definitive treatment, left posterolateral thoracotomy was performed. It was observed that the left lung was destroyed by old pulmonary Tbc and anterior



**Fig. 4.** Microscopy shows cystic structure composed of pancreatic tissue and mature fat tissue on lung mass (H&E,  $\times 200$ ).

mediastinal mass involved the left upper lobe pulmonary artery with subsequent bleeding and intrapulmonary hematoma. The left pneumonectomy with resection of the mass was performed.

Macroscopically, the specimen consisted of  $7 \times 5 \times 2.3$  cm sized mass with fat, fibrous tissue, bone and cartilage with cystic change (Fig. 3). On histologic examination, the tumor disclosed cystic structures composed of mature squamous epithelium, pilosebaceous glands, mature fat tissue, cartilage and bone tissue as well as pancreatic tissue (Fig. 4). Also, there was intrapulmonary hemorrhage with surrounding young and old granulation tissue formation. Histopathological examination confirmed a diagnosis of a ruptured mature cystic teratoma. The patient remains well after surgery.

## DISCUSSION

Approximately 10~12% of primary mediastinal masses are derived from germinal tissues, both in adults and in children[4]. The tumors are felt to develop from germ cell rests which remain in the mediastinum. Believed to arise from germ tissue of varying potential for differentiation that was left behind during embryogenesis, these neoplasms are classified into four main groups: teratoma and teratocarcinoma, seminoma, embryonal-cell carcinoma, and choriocarcinoma[5].

Mediastinal mature teratoma is a benign, slow growing lesion. These tumors are usually well encapsulated and are

composed either of a single large cystic cavity or of several smaller intercommunicating cystic spaces. On histologic examination, mature tissue from ectodermal, mesodermal, and endodermal germ cell layers is typically present. Mature tissue that recapitulates the histology of any human organ can be found in these tumors. However, the ectodermal component (i.e., skin, sebaceous tissue, neural tissue) is usually predominant[2]. Most patients with teratomas have symptoms caused by the tumor, and only about a third are asymptomatic[2]. Usual symptoms are chest pain, cough, and dyspnea. Severe symptoms such as hemoptysis are more commonly associated with ruptured tumors, although patients can be asymptomatic. Tumor rupture is a serious and unique complication associated with mature mediastinal teratomas- possibly related to the production of proteolytic enzymes produced by glandular tissue within the lesion[3]. Alternatively, rapid tumor enlargement may result in ischemic change that leads to rupture[3]. Rupture can occur in up to 36% of all cases[6], most frequently into the lung and bronchial tree, followed by the pleural or pericardial space. Rupture into the lung causes pneumonia, while rupture into the pleural space will produce a chemical pleuritis with a characteristic fat-fluid level. Our case also developed episode of massive hemoptysis associated with ruptured mature cystic teratoma involving pulmonary artery.

The chest radiography typically reveals a well-circumscribed anterior mediastinal mass that often produces into one of the lung fields. In a recent series, teratomas are usually seen as a mass of about 8~10 cm in length[2]. Calcification is present in up to 25% of tumors[7]. On CT they are well defined, thick walled cystic masses containing a variable admixture of densities: fat, water, soft tissue and calcium. CT can be diagnostic in teratomas by revealing pathognomonic findings of fat and calcific densities[8]. Radiographic recognition from plane film examination is not possible unless teeth or skeletal elements are seen. The demonstration of CT findings of ruptured cystic teratoma is important not only for early diagnosis, but also for surgical planning in determining the presence of tumor invasion of adjacent anatomical structures. Although atelectasis or pneumonitis account for most of the associated adjacent lung consolidation, intrapulmonary tumor infiltration is also possible.

The treatment of choice for ruptured teratomas is surgical excision. Surgical removal is sometimes difficult because of the large size of the tumor and the involvement of other structures: pericardium, lung, great vessels, thymus, chest wall, hilar structures, and diaphragm.

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### =국문 초록=

폐동맥 내로 파열되어 응급수술이 필요했던 전종격동의 성숙 낭포성 기형종을 경험하였기에 보고하는 바이다. 39세 여자 환자가 대량객혈을 주소로 내원하여 기관지 동맥 색전술을 시행받았다. 그러나 기관지 동맥 색전술 시행 10일 후 환자는 갑작스런 대량객혈과 함께 의식저하가 생겨 응급 개흉술을 시행하였다. 수술은 좌측 전폐절제술과 전종격동에 있는 종양절제술을 시행하였으며 조직학적 검사 결과 종양은 성숙 상피세포와 피지선, 성숙 지방조직, 연골, 골조직 등으로 구성된 낭포성 구조물로 관찰되었고, 좌 폐동맥으로 침습된 종양의 일부가 파열되어 폐내출혈을 보이고 있었다.

중심 단어 : 1. 기형종  
2. 객혈  
3. 폐