

종격동 종괴로 발현된 자발성 흉선 낭종 출혈 1예

서울특별시 서울의료원 내과

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Spontaneous Thymic Cyst Hemorrhage Manifesting as a Mediastinal Mass

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Thymic cysts are uncommon benign lesions in the anterior mediastinum. We here describe a 55-year-old male with spontaneous thymic cyst hemorrhage manifesting as a rapidly enlarging mediastinal mass that was resected completely with video-assisted thoracoscopic surgery. To the best of our knowledge, this is the first report of a spontaneous thymic cyst hemorrhage in Korea. In cases of rapidly enlarging mediastinal masses, spontaneous thymic cyst hemorrhage should be considered as a differential diagnosis. (Korean J Med 2016;91:62-65)

Keywords: Thymic cyst; Hemorrhages; Mediastinal cyst

INTRODUCTION

Thymic cysts are relatively uncommon lesions, accounting for approximately 3% of all anterior mediastinal masses [1]. The majority of thymic cysts do not grow in short periods of time [2], and a spontaneous thymic cyst hemorrhage is rare, particularly in adults [3]. Spontaneous mediastinal hemorrhage generally presents with symptoms such as chest pain and dyspnea [4]. Here, we report a rare case of asymptomatic spontaneous hemorrhage of a thymic cyst that manifested as a rapidly enlarg-

ing mediastinal mass.

CASE REPORT

A 55-year-old male underwent a chest radiograph during a medical checkup. The chest radiograph revealed double contours in the right cardiac border and the descending thoracic aorta (Fig. 1A). For further evaluation, a chest computed tomography (CT) scan was performed, which revealed a well-defined 1.1-cm sized lung nodule in the left lower lobe. A short-term follow-up

Received: 2016. 1. 31

Revised: 2016. 5. 24

Accepted: 2016. 6. 7

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*The authors have no conflicts of interest to declare.

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was recommended and the patient visited our hospital two months later for examination of the pulmonary nodule.

The patient had neither dyspnea nor chest pain. He had taken aspirin and clopidogrel since being diagnosed with atrial fibrillation, hypertension, and mitral regurgitation six months earlier. His hypertension was well controlled by medication and he did not have a history of coagulopathy or chest trauma. Everything appeared normal on physical examination except that his heartbeat was irregular due to atrial fibrillation. The patient's pulse rate was 84 beats per minute and his blood pressure was 126/80 mmHg.

Hematological examination revealed hemoglobin levels of 16.0 g/dL, a white blood cell count of 7,400 cells/mm³, a platelet count of 149,000 cells/mm³, blood urea nitrogen of 29 mg/dL, creatinine of 1.0 mg/dL, and normal prothrombin time and activated partial thromboplastin time. An electrocardiogram showed atrial fibrillation while a chest radiograph revealed a newly developed mass-like opacity in the left hilar area that was not present on the original chest radiograph taken two months

prior (Fig. 1B). Chest CT scans with contrast enhancement revealed a well-defined mass measuring 10.8 cm x 4 cm in the anterior mediastinum without interval change of the pulmonary nodule (Fig. 2).

The possibility of a malignant tumor in the anterior mediastinum could not be ruled out. Therefore, we performed a percutaneous CT-guided needle biopsy. The specimen consisted of a cyst-like structure with a large amount of blood and minimal fibrous tissue with a bland-looking epithelial lining. However, from the CT findings, we were unable to exclude the possibility of a rapidly growing mediastinal malignant tumor. Thus, we performed video-assisted thoracoscopic surgery to definitely diagnose the mediastinal mass.

On gross macroscopic examination, the mass appeared as an encapsulated multilocular cyst with internal hemorrhage (Fig. 3A). Ash-colored sclerosis and hematoma formation inside the cyst were observed (Fig. 3B). Pathological examination revealed a multilocular thymic cyst filled with numerous red blood cells (Fig. 4A and 4B), thymic tissue with Hassall's corpuscles (Fig.

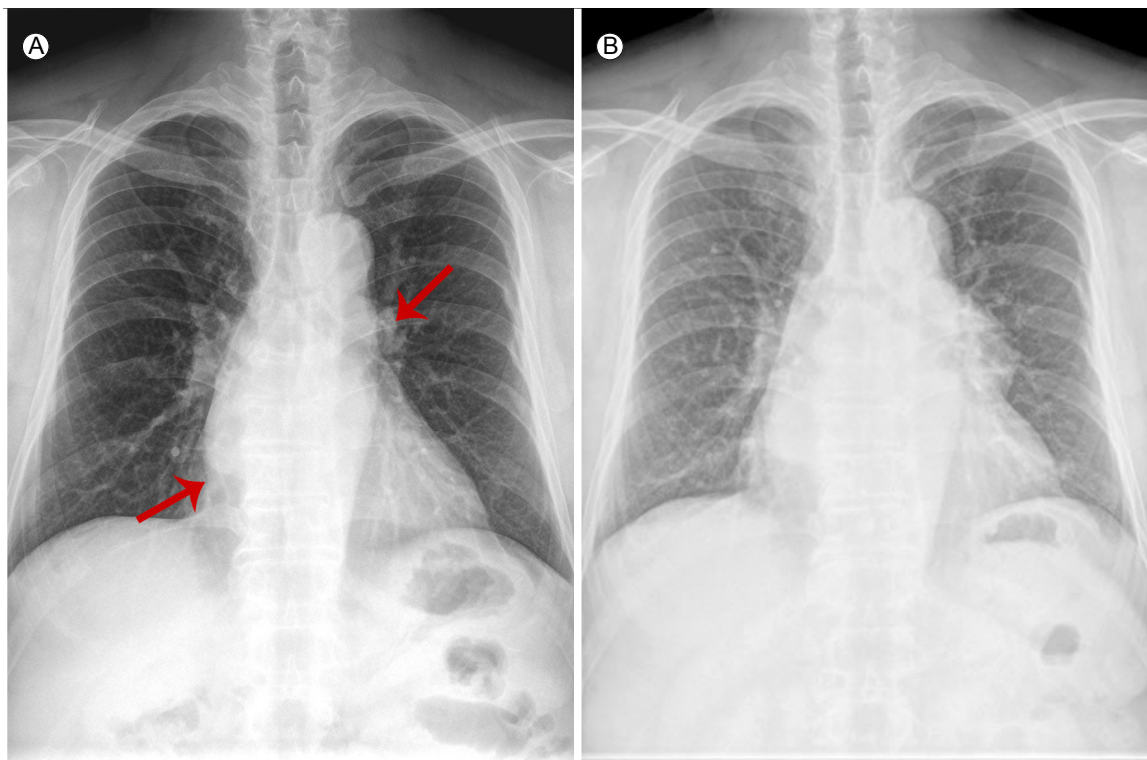


Figure 1. (A) Initial chest radiograph showing double contours in the right cardiac border and the descending thoracic aorta (arrows). (B) Chest radiograph taken two months later showing a newly-developed mass-like opacity in the left hilar area.

4C), and foreign body giant cell reactions, which showed giant cells phagocytizing red blood cells (Fig. 4D). Malignant or atypical cells were not found. We concluded that the rapidly growing mediastinal mass was a benign thymic cyst with spontaneous hemorrhage. The patient was discharged 12 days after the operation without any postoperative complications and was well at

his 5-month follow-up examination.

DISCUSSION

Thymic cysts are benign lesions situated in the anterior mediastinum. They are relatively rare, making up approximately 3% of all anterior mediastinal masses [1]. Thymic cysts are divided into two classes, congenital and acquired [5]. A congenital thymic cyst is comprised of a unilocular cyst with a flimsy translucent wall and serous fluid [1] that is thought to be derived from a persistent thymopharyngeal duct [6]. Thymic tissues adhering to the cyst wall often atrophy without inflammation [5]. On the other hand, an acquired thymic cyst is composed of a multilocular cyst with thick walls and murky fluid related to inflammation [1]. A multilocular thymic cyst can form at any age. It is thought to originate from dilatation of medullary duct epithelial structures involving Hassall's corpuscles, subsequent to an acquired inflammatory process of unknown origin [7]. Reactive lymphoid hyperplasia with germinal centers is frequently found in the cysts, implying that acquired reactive inflammatory processes are involved in the formation of multilocular thymic cysts [5].

Most thymic cysts generally do not grow in a short period of time and have excellent prognoses [2,3]. Nevertheless, a few cases of multilocular thymic cysts are related to thymic neoplasms such as thymoma and thymic carcinoma [8]. Therefore, a thorough sampling of all thymic cyst cases should be performed not only to establish an exact diagnosis but also to rule out the possibility of neoplasm, particularly in cases when the

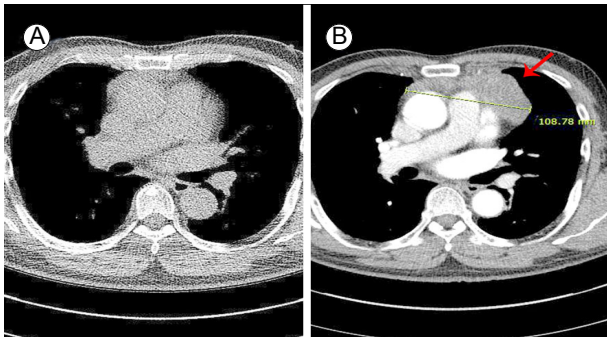


Figure 2. (A) Initial chest computed tomography scans. (B) Chest computed tomography scans with contrast enhancement taken two months later showing a well-defined mass measuring 10.8 cm x 4 cm in the anterior mediastinum (arrow).

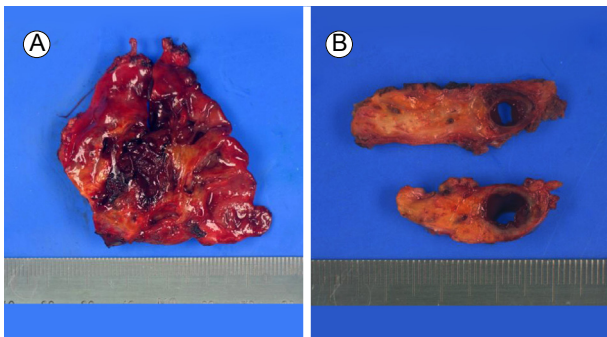


Figure 3. Gross findings. (A) An encapsulated multilocular cyst with internal hemorrhage. (B) Ash-colored sclerosis and hematoma formation inside the cyst.

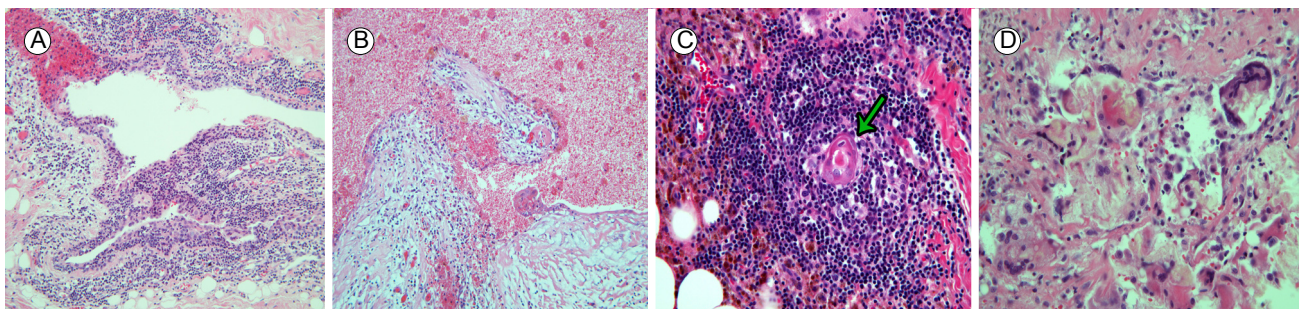


Figure 4. Pathological findings. (A, B) A multilocular thymic cyst filled with numerous red blood cells (hematoxylin & eosin stain, $\times 100$). (C) Thymic tissue with Hassall's corpuscles (arrow, hematoxylin & eosin stain, $\times 400$). (D) Foreign body giant cell reactions showing giant cells phagocytizing red blood cells (hematoxylin & eosin stain, $\times 400$).

wall of the cyst is partially thickened [5].

Spontaneous mediastinal hemorrhage is uncommon. It generally presents with symptoms such as chest pain and dyspnea [4]. Spontaneous mediastinal hemorrhage can occur secondary to the following conditions: 1) complication of a mediastinal mass such as a thymoma, teratoma, malignant germ-cell tumor, retrosternal thyroid mass, or parathyroid adenoma; 2) abrupt sustained hypertension; 3) altered hemostasis as a consequence of uremia [9], hemophilia, hepatic insufficiency, anticoagulant therapy, or thrombolytic therapy; and 4) a transient and steep elevation in intrathoracic pressure during coughing or vomiting. The pathophysiology of spontaneous mediastinal hemorrhage is generally related to the rupture of small mediastinal vessels [3].

In 2002, Nomori et al. [2] reported a case of spontaneous hemorrhage of a unilocular thymic cyst (HTC) that was associated with idiopathic chronic inflammation and presented with chest pain and a rapidly enlarging mediastinal mass. In 2008, Fujiwara et al. [1] reported a case of HTC that was related to thymoma, and also presented with chest pain and a rapidly enlarging mediastinal mass. In 2009, Fernando et al. [4] reported a case of HTC that was linked to thymic carcinoma and presented with chest pain, dyspnea, sweating, and hemoptysis. Like the cases detailed by Fujiwara et al. [1] and Nomori et al. [2], our case presented with a rapidly enlarging mediastinal mass, but symptoms such as chest pain and dyspnea were not observed. Moreover, our case was not associated with underlying causes of mediastinal hemorrhage such as thymoma, thymic carcinoma, or coagulopathy. However, our patient had taken antiplatelet agents, suggesting that these may have provoked hemorrhage of the thymic cyst. Although there has been no previous report of spontaneous hemorrhage of a thymic cyst owing to antiplatelet agents, in our case, the pathophysiology of the spontaneous hemorrhage of the thymic cyst is presumed to be associated with idiopathic inflammation and antiplatelet agents.

Thymic cysts with asymptomatic mediastinal hemorrhage, manifested by a rapidly enlarging mediastinal mass, are extremely rare. To the best of our knowledge, there has been no previous report of spontaneous hemorrhage of a thymic cyst in Korea.

Here, we report a rare case of asymptomatic spontaneous hemorrhage of a thymic cyst manifesting as a rapidly enlarging mediastinal mass in a patient taking antiplatelet agents. The possibility of spontaneous thymic cyst hemorrhage should be considered in the differential diagnosis of a rapidly enlarging mediastinal mass.

중심 단어: 흉선 낭종; 출혈; 종격동 낭종

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