

Surgical Resection of Thoracic Duct Lymphangioma

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A 67-year-old male patient came to the hospital due to lung cancer and mediastinal cystic mass which was suspected to be esophageal duplication cyst. Video-assisted thoracoscopic surgery (VATS) was performed and intra-operative finding suggested it as a cystic mass along the thoracic duct. Thoracic duct was ligated and the cyst was completely resected. A 48-year-old female patient visited the hospital for dysphagia. Mediastinal cystic mass was suspected to be an esophageal duplication cyst. Intraoperative finding suggest a thoracic duct lymphangioma. After thoracic duct ligation, the mass was completely resected with VATS. Postoperative chylothorax did not develop in both cases.

Key words: 1. Thoracic duct
2. Lymphangioma
3. Chylothorax

CASE REPORTS

1) Case 1

A 67-year-old male patient presented to the hospital with a right lower lobe nodule and a paraesophageal cystic mass, which were discovered by regular chest computed tomography (CT). The 2.5-cm right lower lobe nodule was suspected to be a lung cancer. The 4-cm paraesophageal cystic mass was located on the left side of the esophagus (Fig. 1). The mass was suspected to be an esophageal duplication cyst or a bronchogenic cyst.

Video-assisted thoracoscopic surgery (VATS) was performed. The right lower lobe nodule was proven to be adenocarcinoma by a frozen biopsy, and a right lower lobectomy and mediastinal lymph node dissection were carried out. After further dissection through the mediastinum, the mediastinal mass was located on the left side of the esophagus.

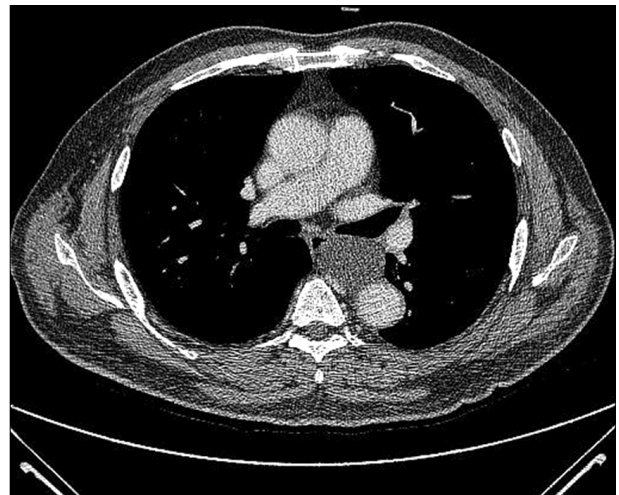


Fig. 1. Case 1. Chest computed tomography shows 4-cm sized mediastinal cystic mass adjacent to left side of the esophagus. The mass was suspected to be an esophageal duplication cyst or a bronchogenic cyst.

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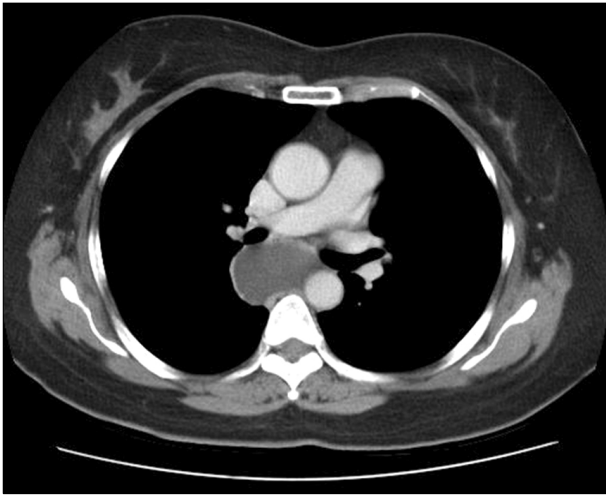


Fig. 2. Case 2. Chest computed tomography shows 5.5-cm sized mediastinal cystic mass which was located on the posterior to the lower esophagus. It was suspected to be an esophageal duplication cyst or a bronchogenic cyst.

However, rather than originating from the esophagus, it seemed that the mass was connected to the thoracic duct. On the basis of this finding, we suspected that the mass was a lymphangioma originating from the thoracic duct. To prevent chylothorax, the thoracic duct was carefully ligated with metal clips and the mass was resected. A 32-French chest tube was inserted into the right pleural cavity.

There was no sign of chylothorax, and the chest tube was removed on the 4th postoperative day. The patient was discharged from the hospital on the 5th postoperative day without complication. The mass was confirmed to be a lymphangioma by a pathologic examination and by immunohistochemistry, on which the mass was positive for CD31, CD34, factor VIII, and D2-40.

2) Case 2

A 48-year-old female patient presented to the hospital for dysphagia that had begun 1 month prior. Chest CT showed a 5.5-cm mediastinal cystic mass, which was located posterior to the lower esophagus (Fig. 2). It was suspected to be an esophageal duplication cyst or a bronchogenic cyst. Endoscopic ultrasonography indicated that it may be a bronchogenic tumor or an esophageal duplication cyst.

An operation was performed by right-side VATS with the

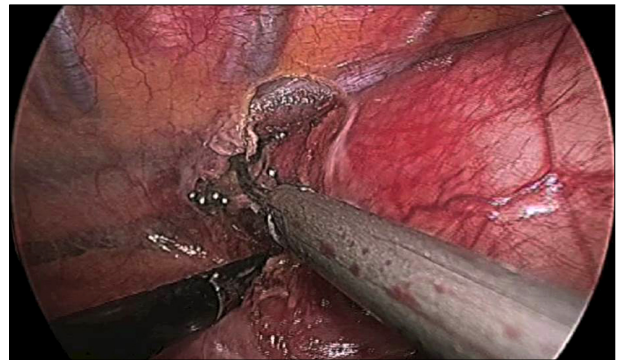


Fig. 3. Case 2. Video-assisted thoracoscopic surgery image shows division of thoracic duct after clipping.



Fig. 4. Case 2. Specimen of 4.5×4.0×1.0-cm sized thoracic duct lymphangioma. The cystic mass was filled with milky fluid and the probe is located in the thoracic duct.

patient in a semi-prone position. The mass seemed to originate from the thoracic duct; therefore, the thoracic duct was clipped, and the mass was excised (Fig. 3). When the mass was dissected on a table, a milky fluid was found (Fig. 4). A 20-French chest tube was inserted into the patient's pleural cavity.

Chylothorax did not occur postoperatively. The chest tube was removed on the 2nd postoperative day. The patient was discharged from the hospital on the 5th postoperative day. In the pathologic examination, the mass was confirmed to be a 4.5×4.0×1.0 cm lymphangioma (Fig. 5). By immunohistochemistry, the mass was demonstrated to be positive for CD31 and negative for D2-40 (Figs. 6, 7).

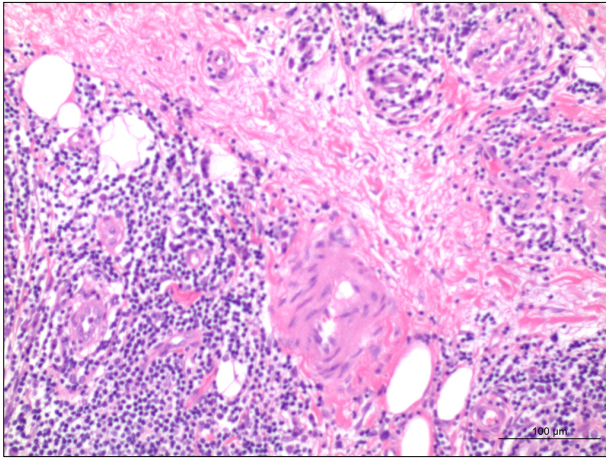


Fig. 5. Case 2. Lymphoid aggregates are observed (H&E, $\times 200$).

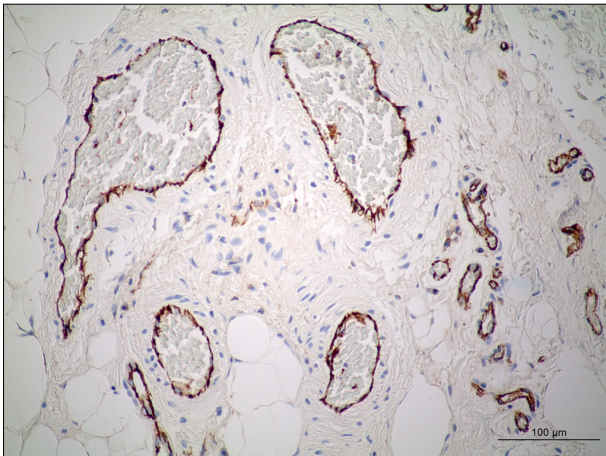


Fig. 6. Case 2. Lymphatic epithelial cells are stained with CD31 antibody (immunohistochemistry with CD31, $\times 200$).

DISCUSSION

The thoracic duct originates in the abdomen at the level of the diaphragm, penetrates through the posterior and superior mediastinum, and exits the thorax through the thoracic aperture, which is usually posterior to the left innominate vein and left of the common carotid artery [1]. Cysts of the thoracic duct can occur either above or below the diaphragm. Supradiaphragmatic thoracic duct cysts are typically found in the neck. However, mediastinal thoracic duct cysts are quite uncommon [2].

The cysts are often symptomatic because of the pressure they exert on the adjacent structures. Symptoms such as

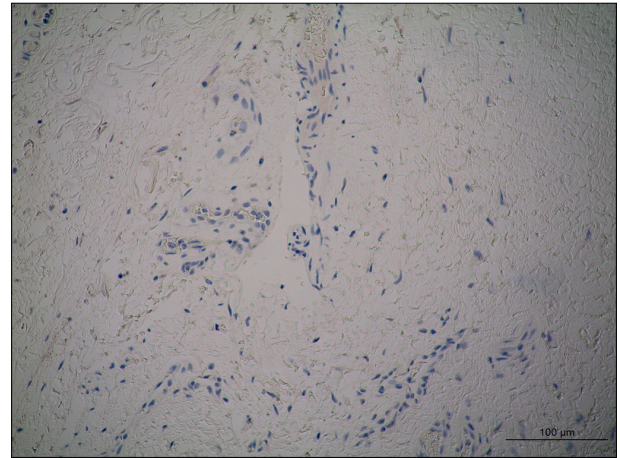


Fig. 7. Case 2. The cystic mass was negative for D2-40 (immunohistochemistry with D2-40, $\times 200$).

coughing, dyspnea, and chest discomfort may be present. Symptoms of dysphagia are often associated with the ingestion of fatty foods. Furthermore, acute respiratory insufficiency after the ingestion of a fatty meal is also observed in some patients [1]. In case 2, the patient experienced dysphagia, which may be caused by the cystic mass.

Bronchogenic and esophageal duplication cysts account for a majority of the mediastinal cysts, and thoracic duct cysts are rarely included in the differential diagnosis. Magnetic resonance imaging is not routinely required for the management of mediastinal cysts but may be able to demonstrate the communication of the cyst with the thoracic duct or the cisterna chyli. The high signal intensity of the T2-weighted images is attributed to the high lipid and protein content in the cyst. Surgical resection of mediastinal cysts, including thoracic duct cysts, is indicated once they are discovered. These cysts are removed to relieve the symptoms and to prevent further growth, infection, and possible malignant transformation [2].

CD-31 and D2-40 are the known markers of lymphangioma [3]. In both cases, the mass was positive for CD-31, which is also called PECAM-1 and is an adhesion molecule that is expressed in the monocytes, granulocytes, and T-cell subsets [4]. D2-40, a monoclonal antibody, which binds lymphatic epithelial cells, stained positive only in the mass from case 1 [5].

Surgical treatment for lymphangioma consists of the removal of the cyst and the ligation of all the lymphatics con-

nected to it [1]. Kwak and Bae [6] reported that they ligated the thoracic duct and that the chylothorax was not observed thereafter. In the case of a mediastinal cystic mass that is adjacent to the esophagus, the mass may originate from the thoracic duct. As such, when removing the mass, the operator should note its relationship with the thoracic duct. If the mass seems to originate from the thoracic duct, it should be ligated to prevent chylothorax.

Here, we report two cases of thoracic duct lymphangioma. Initially, these cases were thought to be esophageal duplication cysts; however, they were found to be thoracic duct lymphangiomas. To prevent postoperative chylothorax, it is important to suspect thoracic duct lymphangioma if a cystic mass is present along the course of the thoracic duct.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Turkyilmaz A, Eroglu A. *A giant thoracic duct cyst: an unusual cause of dysphagia.* J Thorac Cardiovasc Surg 2007; 134:1082-3.
2. Mortman KD. *Mediastinal thoracic duct cyst.* Ann Thorac Surg 2009;88:2006-8.
3. Bhawan J, Silva C, Taungjaruwina WM. *Inconsistent immunohistochemical expression of lymphatic and blood endothelial cell markers in cutaneous lymphangiomas.* J Cutan Pathol 2013;40:801-6.
4. Janeway CA Jr. *Immunobiology: the immune system in health and disease.* 6th ed. New York: Garland Science; 2005.
5. Kahn HJ, Bailey D, Marks A. *Monoclonal antibody D2-40, a new marker of lymphatic endothelium, reacts with Kaposi's sarcoma and a subset of angiosarcomas.* Mod Pathol 2002;15:434-40.
6. Kwak MY, Bae CH. *Thoracic duct cyst in mediastinum: a case report.* Korean J Thorac Cardiovasc Surg 2011;44:83-5.