Successful Excision of an Isolated Mediastinal Cystic Lymphangioma with Bilateral Thoracoscopic Surgery

Masato Kanzaki, MD, Takuma Kikkawa, MD, Tetsuya Obara, MD, PhD, and Takamasa Onuki, MD, PhD

Lymphangioma is a well-known benign tumor and its cystic abnormalities of the lymph vessels are predominantly congenital. Cystic lymphangioma usually occurs in the neck, axillary region, and rarely in the mediastinum, which frequently occurs in children and young adults. A 20-year-old woman had symptoms of palpitation, cough, and dyspnea during the recent 1 month. Both chest computed tomography and magnetic resonance imaging of the chest revealed a well-defined, 13 × 10-cm cystic lesion in the anterior mediastinum. The patient underwent bilateral video-assisted thoracoscopic excision of the cyst and lymphangioma was confirmed based on histopathologic examination. Here, we report a rare case of isolated mediastinal cystic lymphangioma that was successfully excised using a minimally invasive technique.

Keywords: lymphangioma, mediastinal tumor, mediastinum, minimally invasive excision, video-assisted thoracic surgery

Introduction

Lymphangioma is a well-known benign tumor and is cystic abnormalities of the lymph vessels that are predominantly congenital. Most cystic lymphangiomas occur in the primitive lymph sacs of children or young adults. Nearly 90% of cases are diagnosed at approximately 2 years of age.1) Those developing in the mediastinum, which account for less than 1% of cases, are very rare, mostly asymptomatic and usually found accidentally in adulthood.2) Here, we report a rare case of isolated mediastinal cystic lymphangioma that was successfully excised using a minimally invasive technique such as thoracoscopic surgery.

Case Report

A 20-year-old woman had symptoms of palpitation, cough, and dyspnea during the recent 1 month. A chest radiograph showed a large, homogeneous opacity in the left mediastinum with cardiac shadow (Fig. 1). Both chest computed tomography and magnetic resonance imaging of the chest revealed a well-defined 13 × 10-cm cystic lesion in the anterior mediastinum bulding towards both hemithoraces (Fig. 2A–2C). The patient underwent bilateral video-assisted thoracoscopic excision of the multi-loculated cyst (Fig. 3). In particular, thermal injury of phrenic nerve should be avoided. We divided the area between phrenic nerves and the tumor with scissors. There was a large 13 × 10-cm cyst containing serous fluid in the anterior mediastinum intraoperatively, which was no adherent surrounding organs. The histopathologic examination revealed cystic lymphangioma (Fig. 4A and 4B).
Mediastinal Cystic Lymphangioma

Fig. 1 Chest radiograph shows a large, homogeneous opacity in the left mediastinum with cardiac shadow.

Fig. 2 Computed tomography of the chest revealing a well-defined cystic, homogeneous, and low-attenuation lesion in the paracardiac region (A). Magnetic resonance imaging of the chest revealing a mass with heterogenous low signal intensity on enhanced T1-weighted images (B) and high signal intensity on enhanced T2-weighted images (C).

Fig. 3 Intraoperative view shows that a large cyst containing serous fluid in the anterior mediastinum, which was no adherent surrounding organs.

PC: pericardium; LLL: left lower lobe; LUL: left upper lobe

Atypical feature, such as, endothelial tufting, atypia and mitotic activity of lymphatic endothelium were absent. Tumor cells confirmed the lymphatic vessel marker (both D2-40 and CD31) positively (Fig. 4 C and 4D). The 2-year postoperative period was uneventful, and the patient is doing well.

Discussion

Lymphangiomas, also called cystic hygroma, lymphatic or chylous cysts, are rare benign vascular malformations of the lymphatic system composed of cystically dilated lymphatics. Therefore, lymphangioma can occur in any region subserved by the lymphatic system. Although lymphangioma developing in the mediastinum, which account for less than 1% of cases, most lymphangiomas generally appear in the neck (75%) and axillary lesion (20%). Isolated mediastinal cystic lymphangioma are very common and are being more common in the adult age group.1, 2) Thoracic lymphangiomas may remain asymptomatic for years and become apparent with manifestations related to compression of intrathoracic structures, including respiratory symptoms, such as chest pain, cough, dyspnea, vocal cord paralysis, venous
compression, or stridor or dysphagia. Both CT and MRI were helpful in determining the extent of the disease, the cystic, and the lymphatic nature of the mass and can also be used to demonstrate the relationship of the mass and surrounding structures. Treatment of mediastinal lymphangioma remains difficult.\textsuperscript{3-5} Surgical excision is the treatment of choice in cases of cystic mediastinal lymphangioma but complete resection can prove technically problematic because of the insinuating nature of the tumor.\textsuperscript{6,7} However, with complete tumor resection, the prognosis is good. Cases of local recurrence have been reported to be possible if the resection is incomplete.\textsuperscript{8} Other types of adjuvant treatment, such as radiotherapy or injection of sclerosing agent (OK-432), have been proposed; they are still controversial.\textsuperscript{9,10} In conclusion, we advocate thorascopic excision if possible when a mediastinal cystic lymphangioma is suspected.

References