

Multilocular Thymic Cyst with Prominent Lymphoid Follicular Hyperplasia: A Case Report

We present herein an unusual case of multilocular thymic cyst, with prominent lymphoid follicular hyperplasia, in a 64-year-old man. It was incidentally founded as a mediastinal mass on chest radiography, during a routine health check-up. Computed tomography revealed a cystic lesion, which contains thick septa involving the thymus. The resected mass, 8×4 cm in diameter, involved the thymus and there is no adhesion or invasion into the adjacent tissue. The cut surface showed cystic spaces with thick white-tan firm wall, which cysts contained gelatinous material. Microscopically, the lesion was characterized by multiple cysts, lined by flattened cuboidal epithelium that was separated by thick walls, having a dense lymphoid tissue with lymphoid follicles. The patient was discharged without any complication and is well without evidence of recurrence for sixteen months. (**J Lung Cancer** 2012;11(1):45–47)

Key Words: Thymic cyst, Lymphoid hyperplasia, Mediastinal diseases

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Multilocular thymic cyst (MTC) is a rare lesion and consists of multilocular cyst, with thick walls and pericystic fibrosis (1). It is morphologically distinguishable and pathogenetically unrelated to congenital thymic cyst, which is the developmental origin and commonly unilocular, thin-walled. MTCs are thought to be a result of acquired inflammatory process, and have been reported to develop in concert with various mediastinal neoplasms that have intrinsic inflammatory components, such as thymoma, thymic carcinoma, Hodgkin's disease, seminoma, and teratoma (2,3). They are occasionally seen in thymus of myasthenia gravis, human immunodeficiency virus infection and various autoimmune disorders, such as rheumatoid arthritis, systemic lupus erythematosus, Sjögren's syndrome and Hashimoto thyroiditis (4,5). Pseudoepitheliomatous hyperplasia and rim calcification on the wall can be associated with MTC (6,7). MTC is rarely associated with lymphoid follicular hyperplasia (LFH) (5). We, herein, report a rare case of MTC with prominent LFH in a 64-year-old man

with its clinical and pathology features.

CASE REPORT

A 64-year-old man, with a history of atrial fibrillation for 10 years, was incidentally found to have a mediastinal mass on a chest X-ray. He had no subjective symptoms and no abnormalities were noted on the neurological and physical examination. The results of routine laboratory studies were within normal ranges. Chest computed tomography revealed a large cystic lesion, which contains thick septa in the thymus (Fig. 1A). Extended total thymectomy was performed. The mass was oval in shape, 8×4 cm in diameter, and well demarcated from the surrounding thymic adipose tissue, without adhesion or invasion into the adjacent organs. The cut surface showed several cysts, which contains gelatinous material with thick white-tan firm wall (Fig. 1B, C). Microscopically, the lesion was characterized by several cystic spaces, containing

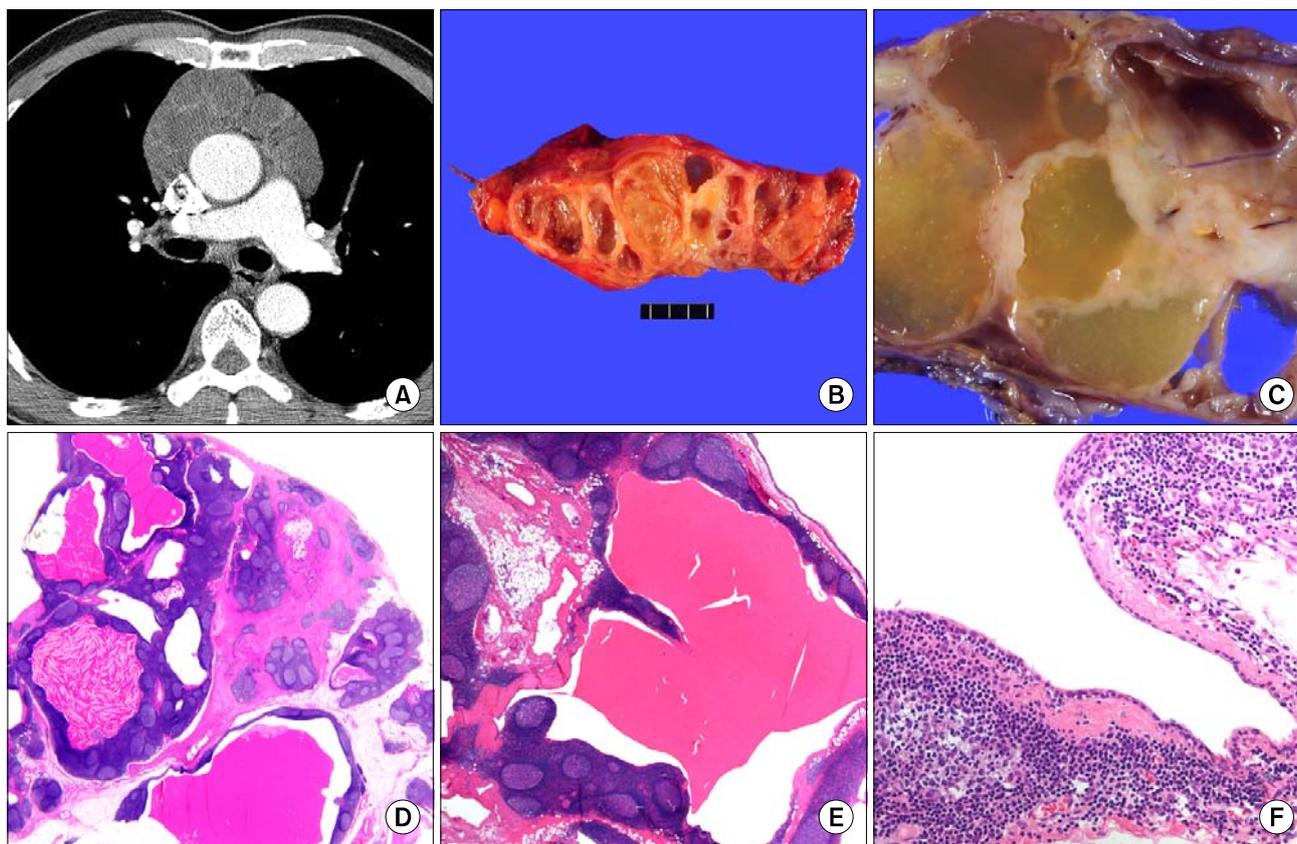


Fig. 1. (A) Chest computed tomography shows a non-enhanced multicystic mass in the anterior mediastinum. (B, C) The mass, 8×4 cm in size, consists of variable sized cystic spaces separated by thick septa. The each cyst is packed with light-yellow gelatinous material and the cyst walls are white-tan in color after formalin fixation. (D~F) Microscopically, the cysts are separated by thick fibrous walls, in which diffuse lymphoid follicle including reactive germinal centers present. The cysts are lined by flattened cuboidal epithelium (D: H&E staining ×100, E: H&E staining ×200, F: H&E staining ×400).

eosinophilic fluid, which spaces are separated from each other by thick fibrous wall and exhibited prominent lymphoid hyperplasia with a well-developed germinal centers (Fig. 1D, E). The cysts are lined by flattened cuboidal epithelium (Fig. 1F). There are several small cholesterol granulomas within the lumen of the cyst and its wall. The patient was discharged without any complication, and is well without evidence of recurrence for sixteen months.

DISCUSSION

Thymic cysts are rare diseases that account for 1% to 3% of anterior mediastinal masses (1,5). They are classified into two groups, congenital and acquired. The former group is commonly unilocular and the developmental origin, which usually have atrophied thin wall and lacks inflammation. The

latter group is usually multilocular and having with thick walls. MTC are thought to be result of acquired process and characterized by multiloculation, thick fibrous walls, and coexistence of inflammation (5). Reactive lymphoid hyperplasia with germinal centers is occasionally observed in MTC, suggesting the involvement of acquired reactive inflammatory processes in the development of MTCs (5). Suster and Rosai (1) postulated that MTC most likely results from the cystic dilatation of medullary duct epithelium-derived structures that includes Hassall's corpuscles induced by an acquired inflammatory process. This inflammation is usually idiopathic, but in some cases, specific etiology was identified, such as human immunodeficiency virus infection (4). Cases associated with autoimmune diseases, including Sjögren's syndrome, have been reported (5).

Diffuse LFH in MTC is very unusual, but distinct lesion. We

can encounter only a case among 13 MTCs in Samsung Medical Center for the past 17 years. To our best knowledge, there are only a few reports, regarding clinical and pathological features of MTC with diffuse LFH. Izumi et al. (5) reported 4 cases of MTC with LFH. In their study, 2 patients (50%) were associated with autoimmune disorder, such as Sjogren syndrome, with which the results indicated that inflammation that has been caused by autoimmune diseases may play, in part, an important role in the development of MTC with LFH. In contrast, our case didn't show any evidence of autoimmune disease. However, careful clinical follow-up is required.

On chest computed tomography scan, the lesion is characteristically demonstrated, as a mediastinal mass containing multiple cysts accompanied. However, this radiological finding is not specific for MTCs, indicating the importance of pathological investigation for accurate diagnosis. MTC can be seen in the thymus harboring nodular sclerosis Hodgkin's lymphoma, seminoma, thymomas with cystic change, and cystic lymphangioma (3).

In summary, we report MTC with diffuse LFH, a rare but distinct lesion. Pathological findings were essential to make the diagnosis, and careful histologic examination should be carried

out to exclude coexisting lesions.

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