

Multilocular Thymic Cyst Associated with Mediastinal Teratoma: A Case Report¹

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Multilocular thymic cyst (MTC) has been reported to develop in concert with various mediastinal neoplasms that have intrinsic inflammatory components, such as thymoma, thymic carcinoma, Hodgkin's disease, and seminoma. However, development of mediastinal teratoma without intrinsic inflammation in association with MTC has rarely been reported. Here, we report the findings of a case of MTC associated with mediastinal mature cystic teratoma on computed tomography (CT) with CT-histopathologic correlation.

Index words : Mediastinum
Neoplasms, CT
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Teratoma

Benign thymic cysts can be congenital or acquired. Congenital cysts are typically unilocular and thin-walled to the point of translucency; careful histopathologic examination shows no evidence of inflammation. In contrast, acquired thymic cysts are usually multilocular, hence the commonly used term "multilocular thymic cyst (MTC)." These cysts have thick, fibrous walls, and histopathologic examination typically reveals significant inflammation and fibrosis (1 - 3).

Multilocular thymic cyst can develop in concert with various mediastinal neoplasms including thymoma, thymic carcinoma, Hodgkin's disease, and seminoma (1, 2). Two cases of MTCs associated with benign mediastinal teratomas have been reported, but to our knowledge, detailed CT features and CT-histopathologic correlations have not been described (1). Here, we report the

findings of a case of MTC associated with mediastinal mature cystic teratoma on computed tomography (CT) with CT-histopathologic correlations.

Case Report

A 13-year-old boy presented with a 2-month history of chest pain. He had no history of previous surgical procedures or malignancy. A human immunodeficiency virus (HIV) test was negative. Chest CT revealed a well-circumscribed lobulated mass predominantly in the left anterior mediastinum that measured about 8.0 × 5.3 × 12.0 cm in size. Contrast enhancement revealed a heterogeneous mass, mainly consisting of multilocular cystic portions with enhancing intervening septae and some solid areas (Fig. 1). The upper portion of the mass extended to just below the thyroid gland and showed multiple small cysts separated by fine, enhancing septae. The lower portion of the mass consisted of two cystic areas between which small calcific dots were noted. Normal thymic tissue was present to the right lateral of the lower cystic portion. A small area of fat attenuation

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was observed in the lower left portion of the mass (Figs. 1B and 1C). Minimal atelectatic changes were noted in the left upper lung, which contacted the mass. There was a small amount of pleural effusion in the left hemithorax.

Based on the radiological diagnosis of a ruptured teratoma, the mediastinal mass and adjacent normal thymus

was resected; on gross examination, two distinct areas were found within the mass. The first area, which corresponded to the left lower cystic portion on CT, contained a mixture of solid and cystic components. The cysts were filled with sebaceous material and dirty fluid. Fat tissue, corresponding to fat attenuation on CT, was found along the outer cyst wall. Adhesion between the



Fig. 1. A 13-year-old boy with coexistent acquired multilocular thymic cyst and mature cystic teratoma in the anterior mediastinum.

A-C. Axial scans (a and b) and coronal reformat image (c) show a lobulated mass in the anterior mediastinum that extends to just below the thyroid gland and demonstrates heterogeneous enhancement, mainly consisting of multilocular cystic areas. Note multiple thin septae (arrowheads in a and c) and areas of fat attenuation (solid arrows in b and c) within the mass. Upon CT-histopathological correlation, atelectasis (empty arrow in c) in the lung parenchyma abutting the mass and left pleural effusion (not shown) suggested teratoma rupture. The boundary between the multilocular thymic cyst and teratoma is indicated by the dotted lines in b and c.

D. Gross specimen of the mediastinal mass shows mixed areas of solid and cystic portions. The normal thymus (Th) is seen on the right side of the mass, the collapsed multilocular thymic cyst (Cy) in the middle, and teratoma (Te) on the left side. Note the septae within the collapsed thymic cyst that give it a multilocular appearance. The apex of the mass (arrow) that extended to the thyroid gland on CT was a portion of the multilocular thymic cyst. The region of teratoma rupture (curved arrow) is also noted on the left upper portion of the gross specimen. Arrowheads indicate fat components within the teratoma that correspond to areas of fat attenuation on CT. The boundary between the multilocular thymic cyst and teratoma is indicated by the dotted line.

apical portion of the cyst and the abutting lung parenchyma had features suggestive of rupture; this adhesion corresponded to the atelectatic changes peripheral to the mass on CT. Histologic sections of this first area revealed mature cystic teratoma. Chronic inflammatory changes were noted only at the site of rupture.

The second area was composed of normal thymic tissue and multiple cysts filled with serous fluid. The cysts corresponded to the upper multilocular and right lower cystic areas of the mass on CT. The sections from the multicystic area revealed pronounced acute and chronic inflammatory responses and old hemorrhage, which are characteristic of MTCs. A histological diagnosis of MTC in association with ruptured teratoma was rendered based on these findings.

Discussion

Multilocular thymic cysts may develop de novo or be associated with a variety of systemic and localized mediastinal disorders. The latter include HIV infection, seminoma, yolk sac tumor, lymphoma, thymoma, thymic carcinoma, Langerhans cell histiocytosis, Sjögren's syndrome, myasthenia gravis, systemic lupus erythematosus, and prior thoracic surgery or irradiation (1 - 3). After reviewing the clinical and pathologic features of 18 cases of MTC, Suster and Rosai (3) concluded that the formation of multiple cysts was a secondary reaction induced by an "idiopathic" inflammatory process within the thymus. They also postulated that MTCs associated with tumors were induced by an inflammatory component within the tumor rather than by the mass effect of the tumor itself. Thus, they argued that MTCs would not be associated with thymic tumors that lack intrinsic inflammatory components, such as teratocarcinoma (3).

Contrary to Suster and Rosai's argument (3), MTC was associated with a benign cystic teratoma in this case, and to our knowledge, in two other such cases reported in the literature (1). As expected in such tumors, the associated benign cystic teratomas in these three cases did not harbor any intrinsic inflammatory components. Although the current case exhibited limited chronic inflammatory changes, these were found only at the site of teratoma rupture, apart from the MTC area. Rakheja et al. (1) presumed that the associated teratomas might not have been incidental, but rather were the inciting agents that induced cyst formation and inflammation of MTC.

On CT, MTCs appear as well-defined, heterogeneous-

ly-enhancing, unilocular or multilocular cystic masses arising at the location of the thymus, often with calcification or soft-tissue attenuation components (2). The radiologic differential diagnosis for such lesions in the anterior mediastinum includes cystic teratoma, lymphangioma, hemangioma, cystic degeneration of seminoma, Hodgkin's disease, and cystic thymoma. Among these, cystic teratoma is the most difficult to confidently differentiate from MTC because it shares common radiological features with MTC, including a unilocular or multilocular cystic mass and frequent calcifications. Identification of cartilaginous or tooth-like calcifications or fat in the lesion suggests teratoma, as in this case. Because of the radiological similarities, accurate distinction between areas of MTC and teratoma on CT is possible only after CT-histopathological correlation.

Radiological diagnosis of MTC associated with teratoma is very difficult because of their radiological similarity. Thus, MTC associated with teratoma should also be considered along with the aforementioned diseases in a patient with a multilocular cystic mass in the anterior mediastinum, particularly because MTC may recur after excision and adhere to adjacent structures, simulating an invasive neoplasm at thoracotomy (2, 3).

Mediastinal teratomas may infrequently rupture into the adjacent structures, such as the pleural space, pericardium, lung parenchyma, or tracheobronchial tree. CT findings of internal inhomogeneity within the mediastinal teratoma and changes in the adjacent lung parenchyma, pleura, or pericardium are possible signs of tumor rupture (4). In our case, ipsilateral pleural effusion and atelectatic changes abutting the mass suggested tumoral rupture. Proteolytic or digestive enzymes derived from the tumor are the proposed cause of tumoral rupture (5).

In summary, we report a case of MTC associated with mature mediastinal teratoma. Accurate distinction between areas of the two conditions on CT was very difficult because of their radiological similarity. However, MTC associated with teratoma should be included in the differential diagnosis of a multilocular cystic mass in the anterior mediastinum.

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