Acta Med. Okayama, 2020
Vol. 74, No. 5, pp. 431-433

Acta Medica Okayama
http://escholarship.lib.okayama-u.ac.jp/amo/

Case Report

A Giant Thymic Cyst Accompanied by Acute Mediastinitis

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We encountered a rare case of thymic cyst accompanied by mediastinitis. A 39-year-old Japanese male presented with fever and chest pain. The chest CT revealed a mass composed of a lobular cystic lesion with inflammation, suggesting the onset of mediastinitis. A definitive histological diagnosis was not obtained, and we performed a thymectomy. Pathologically, the thymic cyst was accompanied by multiple cavities, mimicking thymic cysts, caused by the inflammatory abscess. The surrounding adipose tissue showed inflammatory cell infiltrations with chronic fibrosis. These findings indicate that clinicians should be aware that thymic cysts may cause severe mediastinitis.

Key words: thymic cyst, multilocular thymic cyst, mediastinitis

Thymic cysts are rare, comprising 1-5% of mediastinal tumors [1,2]. They are usually asymptomatic and develop mainly in the anterior mediastinum. Thymic cysts are divided into two types: unilocular cyst, which occurs in congenital cases, and multilocular thymic cyst (MTC), which is an acquired disease. Here, we report a rare case of a giant thymic cyst suspected to be MTC accompanied by mediastinitis.

Case Report

A 39-year-old Japanese male was referred to our hospital because of fever and anterior chest pain after a contrast-enhanced computed tomography (CT) scan for an arterial aneurysm in his right hand. The chest radiographs revealed a mediastinal shadow that was not detected by an X-ray examination conducted 3 years before the present onset of symptoms (Fig. 1A, B). The laboratory data obtained at the patient's initial visit to our hospital showed acute inflammation (white blood cell count: 15,640/μl, C-reactive protein: 27.08 mg/dl). Contrast-enhanced CT of the chest revealed that the shadow was a lobular cystic mass with inflammation, suggesting that the mass was accompanied by acute mediastinitis. The mass consisted of multiple cystic lesions measuring 79×60×58 mm (Fig. 2A, B). The findings of mediastinitis were not observed on the non-enhanced CT that was conducted at the time of the examination of an arterial aneurysm of the patient's hand 2 days earlier (Fig. 2C). No allergic symptoms were observed during the contrast-enhanced CT scan.

We performed positron emission tomography-CT (PET-CT) and chest magnetic resonance imaging (MRI). The PET-CT revealed that the solid part of the mass showed areas of heterogeneous ^{18}F-fluorodeoxy glucose uptake. The MRI revealed that the mass was

Received March 24, 2020; accepted June 16, 2020.
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Conflict of Interest Disclosures: No potential conflict of interest relevant to this article was reported.

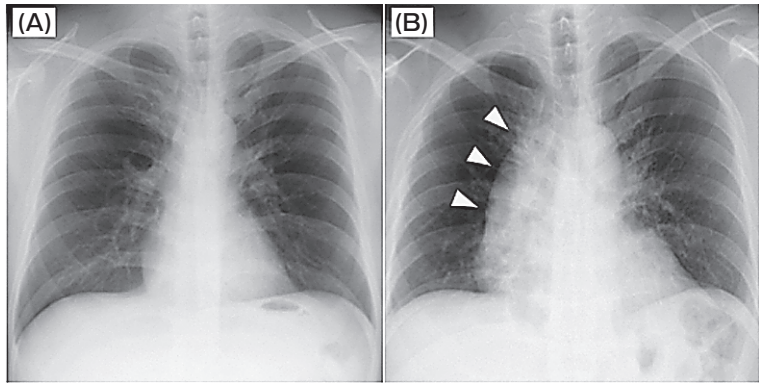

Fig. 1 Chest X-ray (**A**) 3 years before the onset and (**B**) at the time of the patient's present hospitalization. The right mediastinal shadow was enlarged (*arrowhead*).

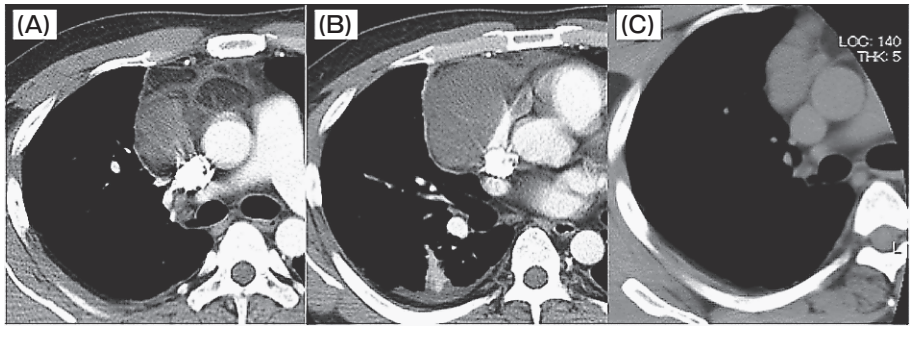

Fig. 2 Enhanced chest CT shows a lobular cystic mass. **A**, **B**, Images at the hospitalization; **C**, There were no inflammatory findings around the mediastinal tumor 2 days prior to the hospitalization.

composed of cysts, with a solid component and adipose tissue. We performed a CT-guided percutaneous needle biopsy of the solid part of the mass, but a definitive histological diagnosis was not obtained. After the procedure, the symptoms of chest pain, high-grade fever, and unilateral pleural effusion worsened. An analysis of the pleural effusion revealed that the amylase level was normal, and there were no findings of malignant cells or bacterial infection.

We then performed an open thymectomy. The mediastinal mass was located at the right lobe of the thymus and had a diameter of 70 mm. The mass showed inflammatory adhesions, but invasion into the adjacent organs was not observed. The mass was composed of a multilocular cystic lesion (Fig. 3A), with gray and bloody fluid. The microscopic findings of the resected specimen are shown in Figure 3B-D; they revealed that part of the cystic lesion was lined by ciliated columnar epithelium (Fig. 3B), although most of the cystic lesion had no lined epithelium. The cystic lesion was characterized by a unilocular thymic cyst, and most of the multiple cysts seemed to be the cavities caused by the inflammatory abscess. The surrounding adipose tissue showed inflammatory cell infiltrations, chronic fibrosis, and abscess (Fig. 3C, D). There were no findings of malignancy or bacterial infections in resected specimen. There has been no tumor recurrence in the 3-year follow-up period.

Discussion

Congenital thymic cysts are typically unilocular. They are derived from the third and fourth branchial pouches [2]. A smooth fibrous capsule lined by epithelium characterizes these cysts. In contrast, acquired thymic cysts are usually multilocular lesions induced by tumors or an inflammatory process [3-5]. They are called MTCs, and they usually appear as soft, rubbery masses with thick and fibrous walls, and the cysts are filled with gray-brown or dark bloody fluid [6]. It is

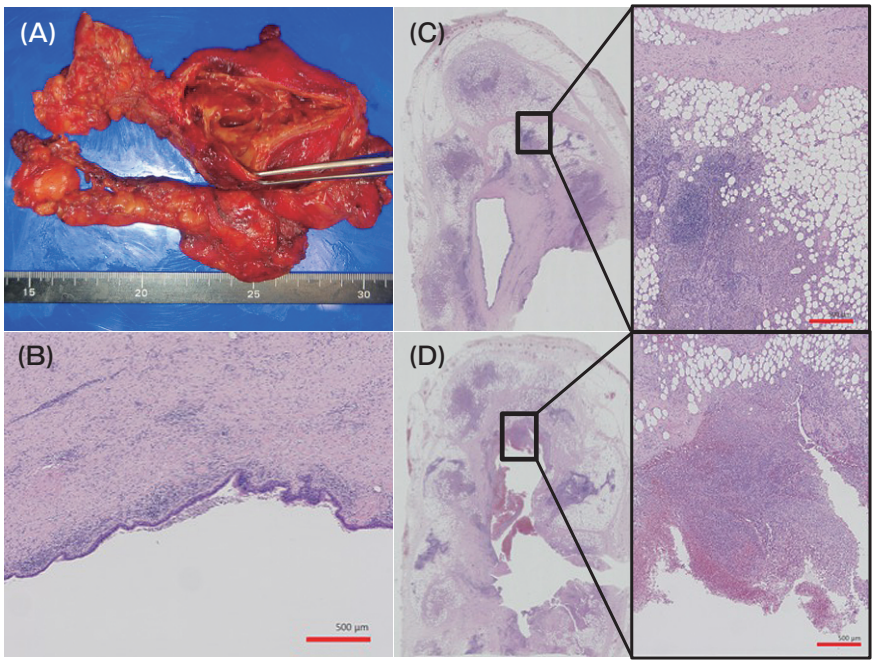

Fig. 3 **A**, Macroscopic findings of the resected specimen showed a multilocular cyst. **B-D**: Histopathological findings (HE staining); **B**, The unilocular thymic cyst is shown; **C**, Fibrosis was observed within inflamed adipose tissues; **D**, Most of the cystic lesion was composed of abscess.

important to differentiate an MTC from a unilocular cyst because MTCs sometimes occur in association with a tumor, infection, or autoimmune disease [6]. Our patient did not have a past history or symptoms suggesting these diseases (including collagen disease). In addition, the resected specimen did not contain a malignant component.

The clinical features of the mediastinal tumor of our patient are similar to those of MTC. We could not confirm the association between mediastinitis and contrast material, and we have found no report showing that an intravenous administration of contrast reagent induced mediastinitis. However, mediastinitis was induced 2 days after our patient's examination, and we speculate that stimulation by the contrast reagent may have triggered the mediastinitis. The existence of the unilocular cyst and adipose fibrosis suggests that the unilocular thymic cyst existed inherently and gradually enlarged over a 3-year period by chronic inflammation from an unknown cause.

Although the process of MTC formation has not been described to date, we strongly suspect that abscesses caused by inflammation took place in a part of our patient's thymus, resulting in the formation of the MTC. Clinicians should be aware that thymic cysts sometimes cause severe mediastinitis, and appropriate management including periodic imaging examinations is important to prevent serious clinical situations affecting the patient.

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