CASE REPORT

Spontaneous Rupture of Idiopathic Thymic Abscess with a Markedly Increased CA-125 Level

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Abstract

We report a rare case of spontaneous rupture of idiopathic thymic abscess into the pleural cavity. A 64-year-old woman was admitted to hospital with pleuritic retrosternal chest pain. Chest roentgenograms disclosed a small amount of bilateral pleural effusion, the examination of which exposed a sterile serous exudate with a markedly increased CA-125 level. Chest computed tomography revealed a large anterior mediastinal cystic mass with bilateral pleural effusions. Following complete resection of the mass, the histological examination revealed cavitary lesion with necrotic thymic tissue and inflammatory infiltrate surrounded by fibrous wall. The immunohistochemical staining for CA-125 displayed strong positivity at the Hassall’s corpuscles. Cyst fluid also revealed a highly elevated CA-125 level. Her serum CA-125 concentration two months after surgery had fallen to 28 IU/L. She is now doing well without recurrence of the cyst five months after surgery.

Key words: thymic abscess, pleural effusion, CA-125, mediastinum


Introduction

Thymic abscess is a very rare disease entity in a geriatric population. We describe a patient who presented with pleuritic chest pain and was found to have an anterior mediastinal mass with bilateral pleural effusion. The patient underwent median sternotomy and complete resection of the mass and the diagnosis of rupture of idiopathic thymic abscess was confirmed by histopathologic examination.

Case Report

A 64-year-old woman with unremarkable medical history was admitted to our institution for evaluation of acute onset retrosternal chest pain. Over the previous two days she had noted a sense of chill associated with pleuritic retrosternal chest pain, which was aggravated while bending forward. She had no history of diabetes, tuberculosis and chest surgery. On admission, she was afebrile, with a body temperature of 36.5°C, respiratory rate of 20/min, and pulse rate of 80 beats/min. She had decreased breath sounds in right lower lung fields. There was no organomegalgy or lymphadenopathy. In laboratory findings, she had a white cell count of 11.9×10^9/L, C-reactive protein level of 0.6 mg/dL (0-0.8 mg/dL), erythrocyte sedimentation rate of 6 mm/hr (0-40 mm/hr) and a mildly increased serum cancer antigen (CA)-125 antigen level of 71.1 IU/L (0-35 IU/L). Electrocardiogram showed normal sinus rhythm without any suggestion of acute myocardial infarction. Her CA 19-9, carcinoembryonic antigen, and alpha-fetoprotein concentrations were within the normal range. Admission blood cultures were negative. A chest X-ray revealed obscuration of upper anterior chest space with small amount of bilateral pleural effu-

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Figure 1. A chest X-ray revealed obscuration of the upper anterior chest space (arrow) with a small amount of bilateral pleural effusions. (a, posteroanterior view; b, left lateral view; c, right decubitus view; d, left decubitus view)

Diagnostic thoracentesis revealed a serous exudative pleural effusion containing white blood cells 1,540/mm$^3$ (lymphocyte 29%, large cell 71%), red blood cells 4,900/mm$^3$, pH 8.0, glucose 115 mg/dl, protein 4.0 g/dl, lactate dehydrogenase 4,860 IU/L, adenosine deaminase 62.6 IU/L, and markedly increased CA-125 antigen level of 2,880 IU/L. The cytological examination of the pleural fluid showed some atypical large cell but absence of any frank malignant cell. A Gram-stained smear and culture of the pleural fluid failed to yield any organism. Test for acid-fast bacillus and PCR for tuberculosis were also negative. Abdominal and pelvic sonogram showed no obvious abdominal and pelvic mass or ascites. Computed tomographic scan of chest showed a large anterior mediastinal dumbbell-shaped cystic mass of 34x23x68 mm size and bilateral pleural effusions (Fig. 2). Although the pleuritic chest pain was diminished with oral analgesics, we performed a median sternotomy and resection of the mass to confirm the true nature of the mass. An ovoid, well-encapsulated, anterior mediastinal cystic mass, which was partially adhered to the innominate vein and pericardium, was completely resected with a part of the pericardium. Yellowish cyst fluid which was aspirated intraoperatively also revealed highly elevated CA-125 concentration of 1,217 IU/L. CA 19-9, carcinoembryonic antigen, and alpha-fetoprotein concentrations in the cystic fluid were within the normal range. Gross and microscopic examination revealed cavitary lesion with necrotic thymic tissue and inflammatory infiltrates surrounded by fibrous wall (Fig. 3). It showed no epithelial lining cells. The surrounding tissue showed chronic active inflammatory process as with residual thymic tissue. The immunohistochemical staining for CA-125 displayed scattered positive

sions (Fig. 1).
Figure 2. Chest computed tomography revealed a large anterior mediastinal dumbbell-shaped cystic mass (arrow) and bilateral pleural effusions. (a, horizontal view; b, sagittal view)

Figure 3. Gross section of the cystic mass revealed several cavities with yellowish necrotic material within the cavity. The periphery of the cavity shows linear congestion and hemorrhage. The surrounding tissue shows dense fibrosis and adipose tissue.

Figure 4. Microscopically, intracavitary necrotic tissue and inflammatory cells are disclosed (a). The surrounding fibrous tissue shows chronic active inflammation and residual thymic tissue characterized by Hassall’s corpuscle (arrow) (b). The immunohistochemical staining for CA 125 reveals scattered positive foci within the abscess cavity as with strong positivity at the Hassall’s corpuscles (c, d). (a and b, hematoxylin and eosin staining, magnification ×100; c and d, ABC method ×200).

foci within the necrotic tissue and strong positivity at the Hassall’s corpuscles (Fig. 4). Acid-fast stain, Gomori methenamine silver stain and periodic acid-Schiff stains were negative for organisms. Culture from the abscess revealed no organism. From these pathological findings, this patient was pathologically diagnosed with rupture of idi-
pathic thymic abscess with chronic mediastinitis. After an uneventful postoperative period, she was discharged. Her serum CA-125 concentration two months after surgery had fallen to 28 IU/L (0-35 IU/L). She is now doing well without recurrence of the cyst five months after surgery.

**Discussion**

We herein present a very rare case of thymic abscess the cause of which was not clinically apparent. This case is of particular interest as it had findings which differed from the cases reported previously. First, this is a case report of thymic abscess in a geriatric patient with no evidence of known predisposing factor such as surgery, trauma or spread of infection from an adjacent region and without bacteremia. In the English language medical literature, there was only one case report in a geriatric patient in which a thymic abscess occurred with staphylococcal bacteremia and had focus of spread from adjacent manubriosternal pyarthrosis (1).

Secondly, in the present case, the CA-125 concentration in pleural and cystic fluid was markedly increased. CA-125 is widely used in monitoring ovarian carcinoma but elevated levels of CA-125 can be due to non-specific stimulation of pleural mesothelium and diverse etiologies (2). In this case, the epithelial cells of Hassall’s corpuscle strongly expressed CA-125. Thymic epithelial cells are thought to produce various cytokines in response to inflammation (3). Because mediators of inflammation can induce CA-125 production in normal epithelium, stimulated thymic epithelial cells are potential sources of CA-125. Serum CA-125 is reported to be elevated in neoplastic thymic cells such as thymic carcinoma or thymoma (4, 5). To our knowledge, the production of CA-125 in benign thymic disease has not been reported before. Therefore, this rare condition needed to be ruled out before considering malignancy and should be considered in the differential diagnosis in postmenopausal patients with mediastinal mass, pleural effusions and elevated serum and pleural CA-125 concentration as in this case.

Patients with an anterior mediastinal mass pose a diagnostic challenge. Differential diagnoses include Hodgkin’s disease, thymic neoplasms, substernal thyroid mass, teratoma and germinal tumor (6). Cystic degeneration of mediastinal tumors, such as thymomas, lymphomas, germ cell tumors, mediastinal carcinomas, has been reported previously (7). Therefore, complete surgical excision should be performed for both diagnostic and therapeutic reasons especially in an elderly patient such as the present case. The presence of Hassall’s corpuscles in the cystic wall in this case may be pathognomonic of an abscess of thymic tissue origin.

**References**