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Case report - Thoracic oncologic

Difficulties in diagnosis and treatment of thymic adenocarcinoma producing β-human chorionic gonadotropin in anterior mediastinum

Yong Joon Ra[a,*], Mi-Ju Bae[a], Yun Seong Kim[b], Kyung Un Choi[c]

[a]*Department of Thoracic and Cardiovascular Surgery, Pusan National University Yangsan Hospital, Mulgum-eup, Beomeo-ri, Yangsan, Kyeongsangnamdo, Korea*
[b]*Department of Internal Medicine, Pusan National University Yangsan Hospital, Mulgum-eup, Beomeo-ri, Yangsan, Kyeongsangnamdo, Korea*
[c]*Department of Pathology, Pusan National University Yangsan Hospital, Mulgum-eup, Beomeo-ri, Yangsan, Kyeongsangnamdo, Korea*

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Abstract

A 68-year-old man with dyspnea and chest pain on left anterior chest during inspiration visited our hospital. An anterior mediastinal mass was detected on a chest computed tomography (CT) and we considered performing surgical treatment. Since the beta-human chorionic gonadotropin (β-hCG) level was increased to 20.46 mIU/ml on the preoperative blood test, incisional biopsy was performed through a Chamberlain incision to rule out the mediastinal germ cell tumors. After diagnosing a benign mass on the postoperative pathological examination of the incisional biopsy specimen, total thymectomy that included the mass was performed via a full sternotomy. On the pathological examination after the second operation, the tumor was diagnosed as thymic adenocarcinoma producing β-hCG, and the tumor had originated from the thymus.

Keywords: Thymic adenocarcinoma; Beta-human chorionic gonadotropin; Anterior mediastinal mass

1. Case

A 68-year-old male patient visited our hospital with complaints of dyspnea and pain on the left anterior chest during inspiration. An anterior mediastinal mass was detected on a chest computed tomography (CT) (Fig. 1), and benign cystic teratoma or thymic tumor's cystic degeneration were suggested for the differential diagnosis. In order to rule the mediastinal germ cell tumors out, the serum tumor markers were measured in serum. The serum tumor markers were measured in order to be distinguished from mediastinal germ cell tumor. The tumor markers, such as lactate dehydrogenase (LDH), alpha-fetoprotein (αFP) were normal. However, the beta-human chorionic gonadotropin (β-hCG) level was slightly increased to 20.46 mIU/ml as compared with the reference range of 0–5 mIU/ml. In order to exclude the probability that the mass would be a mediastinal germ cell tumor, surgical biopsy of the mass was planned and an incisional biopsy of the mass was performed through a left Chamberlain incision. While dissecting the mass for biopsy, it seemed to be a benign mass that did not invade the surrounding structures. When the mass was incised, dirty materials gushed out from inside and the mass looked like fibrotic tissues. A dermoid cyst was suspected due to the gross findings. After the incisional biopsy, it was confirmed that the tissue was fibroadipose tissue with chronic inflammation, therefore, we decided to perform mass excision. Anterior mediastinal mass excision with total thymectomy and mediastinal lymph node dissection were performed via full sternotomy. The patient was discharged without any specific problem at the sixth postoperative day.

For the postoperative gross finding, the cystic portion of the mass, which was caused by necrosis, was in the center of the mass and a solid mass was located in the dependant portion. The mass was capsulated in fibrotic tissue (Fig. 2a). Although the pathological report after incisional biopsy of the mass coincided with the capsule located in the peripheral portion, the solid part of the dependant portion was reported to be adenocarcinoma (Fig. 2a). On pathologic special staining, the tissue strongly stained for mucicarmine (Fig. 2b) and β-hCG (Fig. 2c). After surgery, although abdominal CT and whole body positron emission tomography/computed tomography (PET/CT) were performed to rule out a mediastinal metastasis of unknown primary site, no other primary malignancy sites were found and the mass was diagnosed as a primary thymic adenocarcinoma producing β-hCG.

The patient has been followed up without any specific medical problems.

2. Comment

Anterior mediastinal masses make up more than half of all mediastinal masses and the most of them are of thymic origin. Thymic tumors, such as invasive thymoma and thymic carcinoma, originate from thymic epithelial cells [1–4]. However, thymic adenocarcinoma is a rare histological form of thymic tumor. There have been few

*Corresponding author. Tel.: +82-55-360-2127; fax: +82-55-360-2157.
E-mail address: mdtrimy@gmail.com (Y.J. Ra).

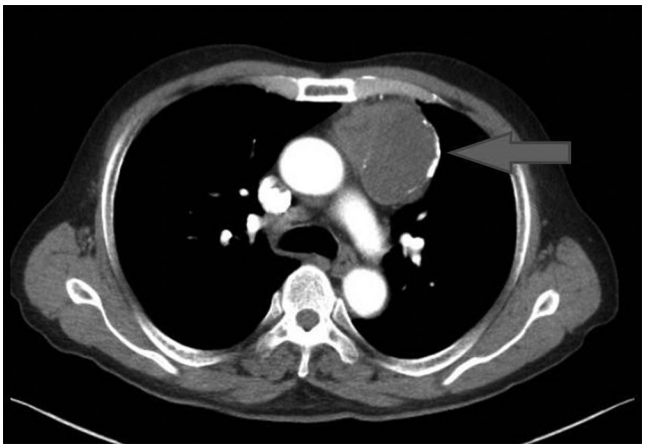

Fig. 1. Preoperative chest CT shows a 6.7 cm sized anterior mediastinal mass. There is calcification in the wall of the mass.

reports that have focused on the clinical prognosis or definite treatment of thymic adenocarcinoma [5], and there only a few reports on resecting thymic adenocarcinoma [5, 6]. An anterior mediastinal mass is generally indicated for surgery at the time of diagnosis because the operative mortality is not high and symptoms can develop due to the mass compressing the surrounding structures whether or not the mass is malignant.

However, in the case of a mediastinal germ cell tumor, chemotherapy or radiotherapy is the primary treatment rather than surgical resection. The tumor markers for distinguishing a germ cell tumor must be measured in all cases when an anterior mediastinal mass is detected. Chemotherapy should be the primary treatment after making the diagnosis via biopsy in the case of increased levels of αFP or β-hCG.

For a seminomatous germ cell tumor [4] that shows an increased level of β-hCG among tumor markers, radiation therapy was the previous initial treatment of choice but cisplatin based chemotherapy has now become the primary treatment. If a remnant mass appears, then the mass should be kept under close observation and radiation therapy should be given after administering a second line chemotherapy if the mass is observed to grow. It has been reported recently that surgical treatment for a germ cell tumor is not an effective treatment [7].

In this present case, as an increased level of β-hCG was observed before surgery, making the differential diagnosis with non-seminomatous germ cell tumor difficult. We failed to make a correct diagnosis for the benign portion of the mass that was biopsied through mediastinotomy. Moreover, in this case, even if preoperative percutaneous needle aspiration (PCNA) had been performed, a correct diagnosis would probably not have been made. When a slight increase in β-hCG is observed in a patient with an anterior mediastinal mass, we believe that detailed evaluations on the characteristics of the radiology should be essential to make a correct diagnosis of the mass.

In addition, in this case, continuous follow-up for local recurrence at the Chamberlain incision site and assessing

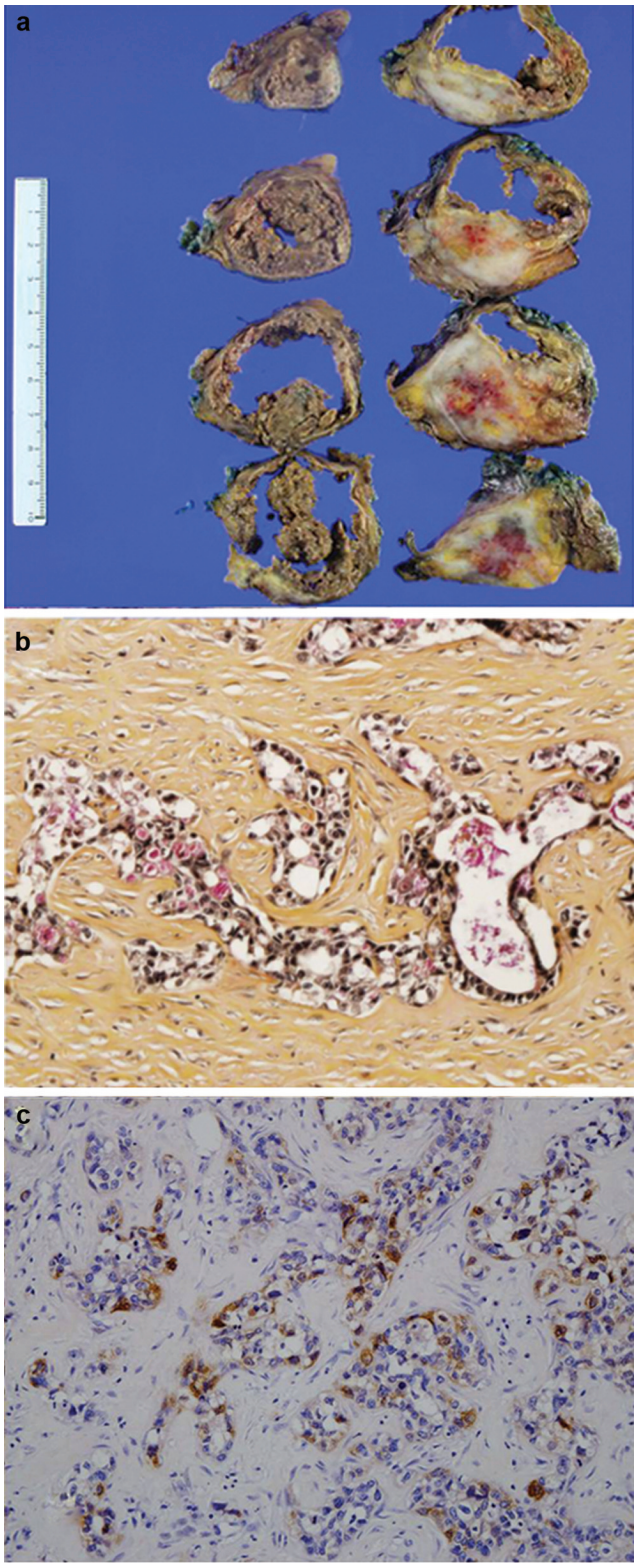

Fig. 2. Pathological features of the mass. (a) Gross specimen of the mediastinal mass. The mass consists of an exterior capsule, a necrotic portion in the center, and the solid portion in the dependant lesion. Incisional biopsy may be carried out in the exterior capsule and the central necrotic portion. The solid portion proved to be an adenocarcinoma. (b) The specimen was stained by mucicarimine. It meant the mass was confirmed as adenocarcinoma. (c) The specimen was stained by β-hCG. It confirmed that this thymic adenocarcinoma produced β-hCG.

the serum β-hCG due to its role for possible tumor recurrence of the mass and a long-term prognosis are required.

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