Thymic Carcinoma which Developed in a Thymic Cyst

Shu-ichi YAMASHITA, Hiroshi YAMAZAKI, Tatsuji KATO, Tada-aki YOKOTA*, Nobuhiro MATSUMOTO** and Shigeru MATSUKURA***

Thymic carcinoma was found in a thymic cyst in a 60-year-old woman who was admitted to our hospital due to cough and pain in her right chest. Chest X-ray showed a huge shadow in the right mediastinum. Chest CT scan showed a cystic tumor with a solid mass. An operation revealed a solid tumor in the thymic cyst. Pathologic diagnosis was squamous cell carcinoma, that contained some glandular tissues. The patient has been in a good condition for two years after the operation. Although the concurrent occurrence of thymic carcinoma and thymic cyst is very rare, it should be added to the differential diagnosis when anterior mediastinal cystic tumor is associated with a solid mass lesion.


Key words: squamous cell carcinoma, thymus, anterior mediastinum

Introduction

Thymic carcinoma is a thymic malignant neoplasm, that tends to be an enlarged tumor in the anterior mediastinum (1). Its pathophysiologic behavior has not been fully understood due to its rare occurrence and consequently thymic carcinoma has not been carefully characterized until recently (2, 3). The signs and symptoms of thymic carcinoma are primarily attributed to the compression towards juxtaposed mediastinal structures (1). Thymic cyst also rarely occurs and its signs and symptoms are apparently due to the compression, resembling thymic carcinoma. Only a few cases of thymic carcinoma developing in a thymic cyst have been reported (4, 5). Thymic carcinoma is thought to originate from thymic epithelium (1). Although the similarity in the origin between thymic carcinoma and thymic cyst is controversial, Leong and Brown (5) have reported a rare case of malignant transformation in a thymic cyst. Here, we present a case of thymic squamous cell carcinoma including glandular proliferation which developed in a large thymic cyst.

Case Report

A 60-year-old female, a non-smoker visited our hospital because of cough and chest pain in the right side. Pain was aggravated when she laid on her right side. She had been experiencing a small amount of hemosputum for a few years. Since a huge abnormal shadow was found in her right mediastinum at the out-patient clinic of our hospital (Fig. 1), she was admitted to our hospital.

Physical examination showed a height of 154 cm, body weight 46 kg, blood pressure 90/60 mmHg and body temperature 36.9°C. There was no evidence of clubbed fingers, cyanosis or heart murmur, although breathing sound was slightly weak in the right chest. No edema was present in her face or in her upper extremities. No lymph node was palpable. Laboratory investigation gave the following results: White blood cell count was 8.0x10³/ml, of which 69% was neutrophils. Red blood cell count was 3.96x10⁶/ml, hemoglobin 10.4 g/dl and hematocrit 32.7%. Serum electrolytes, liver function and kidney function were all in the normal ranges. C-reactive protein was 10.8 mg/dl. Though the serum level of human chorionic gonadotropin (HCG) was slightly high at 1.8 mIU/ml (normal <0.8), it not likely clinically significant. The serum level of alpha-fetoprotein (AFP), anti-acetylcholine receptor antibody, carcinoembryonic antigen (CEA), carbohydrate antigen 19-9 (CA19-9), carbohydrate antigen 125 (CA125) and neuron specific enolase (NSE) were all in the normal ranges. The serum level of squamous cell carcinoma-related antigen (SCC) was as high as 6.7 ng/ml (normal <1.3). There was a huge cystic lesion with an irregular septum and a solid mass in the anterior mediastinum, which was found to extend from the upper limit to the lower limit of her sternum bone in a chest CT scan (Fig. 2). There was no enlargement in lymph nodes. Bronchoscopic observation re-
Yamashita et al

Figure 1. Chest X-ray film on admission suggesting the presence of a huge mass in the right mediastinum.

Figure 2. Chest CT scan on admission showing a cystic mass that contains a solid tumor in the right anterior mediastinum.

Figure 3. The resected cystic tumor. Solid mass lesion is clearly found in the cyst.

revealed that both the right truncus intermedius and the middle lobe bronchus were compressed from the anterior side. Neither bone scan nor gallium scintigram showed any lesion other than the mediastinal mass. No apparent lesion was found in either the abdominal or cranial CT scan. Venogram of the superior vena cava showed no occlusion by the tumor.

In carcinectomy, the tumor (14x7-cm) was found in the right mediastinum. No intact thymic structure was detected in the tumor. Since the tumor adhered to the right upper lobe of lung and to the pericardium, we resected the tumor together with the adjacent structures. The resected lesion was a large cystic tumor which contained a solid mass (Fig. 3). The pathologic examination revealed that the solid mass lesion was squamous cell carcinoma which partly contained a proliferated glandular structure. Most cells in the mass had proliferated and formed stratified nests (Fig. 4a). Though production of keratin was not apparent, there were intercellular bridges (Fig. 4b). A part of the tumor was composed of cells showing obvious glandular elements (Fig. 4c). The cystic lesion was consistent with a thymic cyst.

After the operation, she received neither post operative radiation therapy nor chemotherapy with anti-cancer drugs. A topical recurrence of the tumor on the superior vena cava was found 12 months after the operation. She received radiation therapy of 50 Gy, and the recurrent tumor disappeared. She recovered well and no signs of recurrence of tumor or remote metastasis presented in the 24 month examination following the surgery.

Discussion

Thymic carcinoma has been considered to originate from thymic epithelium (1, 2). A variety of thymic cells are likely potentially capable of changing into a peculiar tumor. All of the types of tumors formed in the thymus had once been referred to
Thymic Carcinoma and Thymic Cyst

Figure 4. Microscopic appearance of the solid mass in the resected lesion. Figure clearly indicates the presence of squamous cell carcinoma. Most cells proliferate to form a striated nest (4A) (HE stain, ×100). There is no apparent production of keratin, though there are intercellular bridges (4B) (HE stain, ×400). It is obvious that the tumor contains some granular proliferating cells (4C) (HE stain, ×100).

as thymomas. However, thymoma should strictly indicate neoplasms of thymic epithelium, since the primary cell type in the thymus is of the epithelial cell. About 10% of thymomas may be malignant, yet they have little or no cytological atypia. Rarities are cytologically malignant tumors which are separated from malignant thymomas by being termed thymic carcinoma. Shimosato et al (2) reported that thymic squamous cell carcinoma could be histologically distinguished from thymoma, squamous cell carcinoma of the lung and other tumors growing in the anterior mediastinum. Although thymic carcinoma tends to behave more actively than invasive malignant thymomas, their appearance in CT scan well resemble each other. It is therefore difficult to distinguish them by clinical signs and symptoms or by imaging analyses such as by CT scan (6). Although a tumorous tissue tends to become a large mass in the anterior mediastinum (1), Ando et al (7) reported an occult tumor presenting malignant pericardial effusion. However, the rareness of thymic carcinoma has been the limiting factor for detailed analysis of its characteristics. Recent diagnostic advances in radiology have succeeded in the identification of the thymic cyst as a tumor in the anterior mediastinum in spite of its rareness (4, 5).

Here, we report the case of thymic carcinoma which developed in a thymic cyst. Only a few reports (4, 5) have described complication of the associated of thymic carcinoma and thymic cyst. The thymic carcinomas of those cases were considered to have derived from thymic cysts. It has been revealed that thymic carcinoma is derived from thymic epithelium. Kodama et al (8) later reported that thymic squamous cell carcinoma also originates from epithelial cells in the medulla of the thymus by using an immunohistochemical technique. On the other hand, Kobayashi et al (4) showed that some types of thymic cysts could be attributed to degenerated Hussal bodies, that were likely derived from medullary epithelial cells. The present case showed development of thymic carcinoma in a thymic cyst, indicating a close relationship between thymic carcinoma and
YAMASHITA et al

thymic cyst in their origin.

The tumor in the present case could be curatively resected, yet topical recurrence on the superior vena cava occurred 12 months after the operation. The patient refused to receive irradiation therapy or anti-cancer chemotherapy immediately after the operation. Recurrent tumor on the superior vena cava successfully disappeared by a 50 Gy-irradiation, and no new recurrence has been observed in the 12 months after the irradiation. Distant metastasis often occurs in patients of thymic carcinoma, even when the primary tumor is totally resected operatively (1–3). It has been demonstrated that thymic carcinoma is active enough to result in an unwilling outcome, and even a tiny tumor has been reported to cause serious pathophysiologic condition of malignant cardiac tamponade (7). Furthermore, thymic carcinoma has been indicated to tend to be a cause of remote metastasis (1). Since neither of anti-cancer chemotherapy nor postoperative irradiation therapy is effective for all histologic types of thymic carcinoma, the necessity of such treatments is now controversial.

We herein reported a case of thymic carcinoma which developed in a thymic cyst. Though this complication is very rare, it is recommended to include thymic carcinoma in the differential diagnosis when an anterior mediastinal cystic mass is found with a solid part inside. Although adequate therapeutic methods are not yet established, further study on this subject is expected to develop clinically useful standards.

References