Pulmonary Epithelioid Hemangioendothelioma Coexistent with Pulmonary Metastasis of Thyroid Cancer

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Abstract

We report a 45-year-old man with epithelioid hemangioendothelioma (EH) and simultaneous pulmonary metastasis of thyroid cancer in his lung. Thyroid cancer, and multiple small nodules in both lungs were noted. He underwent total thyroidectomy followed by radiotherapy with 131I. However, 131I scintigraphy showed poor uptake of radionuclide in the nodules, and the size of the nodules remained unchanged. The diagnostic thoracoscopic biopsy showed two types of nodules, some were positive for thyroglobulin and cytokeratin, and others were reactive for factor VIII. The former nodules were diagnosed as pulmonary metastases of thyroid cancer, and the latter EH.

Key words: multiple neoplasms, lung tumor, thoracoscopic biopsy

Introduction

Pulmonary multiple nodular lesions usually indicate pulmonary metastasis in a cancer patient. Although the presence of pulmonary metastasis is important information to predict the prognosis of a cancer patient, the diagnosis of lung metastasis is often made by radiological study without histological examination.

Epithelioid hemangioendothelioma (EH) is a rare pulmonary neoplasm; it is a unique tumor characterized by an epithelioid or histiocytoid endothelial cell (1, 2). EH usually shows multiple nodular lesions on a chest X-ray (2-4), and it is sometimes difficult to distinguish EH from multiple pulmonary metastasis.

Here, we present a case with multiple lung nodules which consisted of EH and pulmonary metastasis from the thyroid cancer.

Case Report

An asymptomatic 45-year-old man was transferred to our hospital for further evaluation of multiple nodules on a chest X-ray at a health screening in February 1997. On admission, a thyroid nodule of the right lobe, 2x2 cm, and swelling of the cervical lymph nodes were observed. Hematological and biochemical examinations were normal except for a high thyroglobulin level of 121.9 ng/ml (normal <40 ng/ml). Carcinoembryonic antigen and alpha-fetoprotein were also within normal limits. The chest X-ray revealed multiple nodular lesions with irregular borders in both lungs. Chest computed tomography (CT) scans revealed innumerable various sized nodules up to 1 cm in diameter in both lungs (Fig. 1). Based on the pathological study, the thyroid mass was diagnosed as papillary carcinoma, and the multiple lung nodules were considered pulmonary metastases. He underwent total thyroidectomy and cervical lymph node dissection in August 1997, and then, 3,330 MBq 131I radiotherapy was performed. A confirmation study by 131I

Figure 1. Chest CT scans revealed innumerable various sized nodules up to 1 cm in diameter in all areas.
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Figure 2. Microscopic appearance of the small nodules showing adenocarcinoma with papillary growth pattern and some follicular pattern, as well as a positive immunohistochemical reaction for thyroglobulin (A, HE stain, ×100; B, immunostaining with anti-thyroglobulin, ×100).

Figure 3. Microscopic appearance of the larger nodules showed atypical cells with fibrinoid change and hyaline chondroid appearance. The tumor had a gradient of cellularity from a hypocellular sclerotic center bordered by a more cellular periphery. Immunohistochemically, the atypical cells showed a positive reaction for the endothelial marker, factor VIII (A, HE stain, ×15; B, HE stain, ×100; C, Immunostaining with anti-factor VIII, ×100).
scintigraphy, however, showed poor uptake of radionuclide in the nodules, and the size of the nodules remained unchanged against the radiotherapy. A thoracoscopic lung biopsy from the lingula was performed for evaluation of the nodules in April 1999. The thoracoscopic view showed white small nodules on the surface of the lung.

The biopsy specimen, 68x60 mm, included 7 nodules. There were two types: five of them were less than 3 mm in diameter and the others were up to 1 cm in diameter. Macroscopically, the small nodules were white and firm, and did not show calcification or necrosis. The large ones were white and ill-defined and were accompanied by focal pleural thickening. The histological findings of the small nodules showed adenocarcinoma with a papillary growth pattern and in some parts, a follicular pattern, which was an identical finding to thyroid cancer (Fig. 2A). These adenocarcinoma cells were positive for thyroglobulin and cytokeratin (Fig. 2B), and they were considered metastases of the thyroid cancer. On the other hand, the large ones consisted of atypical cells with fibrinoid change and hyaline chondroid appearance (Fig. 3A and B), and factor VIII was positive for the atypical cells (Fig. 3C). The atypical cells were not reactive for cytokeratin or thyroglobulin. The histology of the large nodules was quite different from adenocarcinoma. From these findings, the larger nodules were diagnosed as EH. The patient has survived with no change 18 months postoperatively.

**Discussion**

There are many pulmonary diseases showing multiple nodular lesions including hematogenous metastases, pulmonary arteriovenous malformation, granulomatous infection, Wegener’s granulomatosis, sarcoidosis, non-Hodgkin’s lymphoma, pneumoconiosis, multiple hematomas and so on (5, 6), and they all have different prognoses.

Epithelioid hemangioendothelioma is a rare pulmonary neoplasm characterized by multiple nodular opacities in both lungs (2-4). The prognosis of EH is generally good, and some patients with EH have lived with tumors for more than 15 years (3, 4). However, there were a few patients whose condition rapidly deteriorated, and they died due to respiratory failure (3, 4).

It is fairly rare that multiple small lung nodules consist of two different diseases. Dail et al (4) reported a case with pulmonary EH with primary lung cancer. In their report, the diagnosis was made by histological examination of the surgical specimen. In the present case, pulmonary nodular lesions were recognized as pulmonary metastases of the thyroid cancer only by the radiological findings, and histological examination was not performed before the treatment with 131I. Because of the coexistence of a metastatic lesion, this radiotherapy had acceptable results in this patient. However, we should pay more attention to the possibility of two or more different diseases existing in the lung simultaneously, although the frequency is low.

According to limited information from the radiological study, it is very difficult to distinguish EH from the other pulmonary diseases showing multiple nodular lesions, especially pulmonary metastasis from other organs. Fiberoptic bronchoscopy is usually performed for evaluating multiple nodules in the lung, but it is often difficult to obtain enough information in cases of EH or pulmonary metastasis. Therefore, lung biopsies are sometimes necessary to make an accurate diagnosis of multiple lung nodules.

**References**