Atypical Peripheral (Bronchiolar) Adenoma of the Lung

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An unusual type of peripheral adenoma of the lung in a 52-year-old Japanese female was reported. Clinically the patient showed no sign of hormonal disturbances. Surgically removed tumor was 7 cm in diameter and histologically consisted of ovoid or fusiform cells of spindling and rather bland character, which were arranged prominently in solid nesting pattern suggestive of neurilemmoma or chemodectoma. However, a direct transition from the bronchiolar epithelium to the tumor cell nesting was confirmed by semi-serial sections. In comparison with the usual type of pulmonary adenomata, histological characteristics in this case were discussed in reference to the histogenesis based on constitutional differences between Japanese and Anglo-American races.

Peripheral adenomas of the lung are of particular interest on account of their histological varieties and several unsolved problems in their genesis, compared with usual bronchial adenomas. Since the first description by Brock in 1938, a number of cases have been reported; in the majority of cases the tumors are histologically of carcinoid type, whereas chemodectoma-like or neurilemmoma-like appearance is extremely rare.

In order to approve the histological variety of peripheral adenoma of the lung as a pathological entity, the present paper deals with an unusual type of this tumor with a histological simulation of chemodectoma in some parts and of neurilemmoma in other.

CASE REPORT

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Clinical course. A 52-year-old Japanese female was admitted to the Miyagi Prefectural Hospital on July 9, 1964, because of sore throat, cough and hemoptysis 5–6 times a day. She had no fever or dyspnea. Twenty-eight years before the admission, she had an abortion with hydatidiform mole, and twenty-seven years

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previously, when she had been 25 years old, hysterectomy had been performed because of uterine myoma.

Physical examinations were essentially negative except for slightly suppressed breath sounds over the right back. The hemogram, the ECG and functions of the lung and liver were normal. Urinalysis was negative. There were no malignant cells in gastric juice. In the sputum, leukocytes, streptococci, staphylococci, diplococci and bacilli were found, but no tumor cells and tubercle bacilli were detected.

The roentgenogram revealed a massive spherical shadow about 7 cm in diameter, which was located in the lower field of the right lung adjacent to the mediastinal shadow (Fig. 1). By bronchoscopy and bronchography it was confirmed that the tumor was situated in the middle portion of the lower lobe near the hilum of the right lung (Figs. 2–4). Besides this tumor, a well-demarcated small tumor was found in the anterior mediastinum by the lateral view of the chest, which was interpreted as a cystic tumor of the thymus (Fig. 4).

On August 11, 1964, lobectomy of right middle and lower lobes was performed. The tumor seemed to have arisen in and around the B–9 bronchus. It was compressing the adjacent tissues of middle lobe and sinking in it. Mediastinal, bronchopulmonary and bifurcation lymph nodes were swollen up to 2–3 cm in diameter, but the absence of metastases was confirmed by gross and histological examinations. The tumor in the anterior mediastinum was not removed according to the suggestion of anesthesiologist, general conditions of the patient being regarded as inadequate for further operations. The postoperative course was uneventful.

Before and after the operation, the carcinostatic agents were administered over three months until the time of discharge on December 10, 1964. Follow-up examinations by the chest roentgenography revealed that the tumor-like shadow in the anterior mediastinum was gradually displaced to the right together with the shadow of the mediastinum. On an x-ray film of September 27, 1966, however, the size of the tumor shadow was almost the same as that of pre-operative stage over two years ago.

Pathological findings. On the gross examination of the specimen, a well-circumscribed tumor of 6×6 cm in size was revealed, which was rather soft and elastic, solid and hyperemic. On the cut surface of the tumor there was faintly lobulated texture (Fig. 5). Histologically, the tumor was encapsulated by thin, partly membranous connective tissues, most of which were lined by single cell layer of bronchiolar epithelium. No infiltrative growth was observed anywhere. Most of tumor cells were ovoid or fusiform in appearance. The nuclei of tumor cells varied considerably in shape, i.e., they were round, ovoid, elongated, occasionally swollen and in rare instances also thin and distorted. They had distinct nuclear membranes and relatively high chromatin contents (Figs. 7 and 8). Nucleoli were not prominent and no mitotic figures were found. The cytoplasm was of moderate amount and occasionally contained fine somewhat
Fig. 1. Bronchial adenoma. A simple nodule of 7 cm in diameter is located in the right lower field adjacent to mediastinal shadow.

Fig. 2. The bronchography shows no relationship between nodules and larger bronchi.

Figs. 3 and 4. The planogram appears well demarcated spherical shadow in the middle portion of right lower lobe.
eosinophilic granular elements. The PAS reaction was negative in all of tumor
cells. The Fontana and Hortega stain for argentaffin and argyrophil granules
failed to demonstrate ones. These cells took a solid epithelium-like arrangement,
prominently with a nesting pattern and an acinar structure in the greater part of
the tumor (Fig. 6). There were no distinct glandular cavities, but exceptionally
a few cleft-like spaces of undetermined nature were observed; it was not clear
whether they were true glandular structures or artefacts (Fig. 7).

In some areas, tumor cells were elongated and appeared flattened or spindle-
shaped, and in occasion even fibrillar with palisade-like arrangements suggestive
of neurilemmoma (Fig. 8). In such cases tumor cells were hardly distinguishable
from stromal cells. In general, the stromal cells were very sparse with poor
vascularities. In other areas, peculiar lymphocytic infiltrations were observed
(Fig. 9). In one part of the tumor, a direct transition from the bronchiolar
epithelium to tumor cells was clearly recognized by semiserial sections (Fig. 10).
The tumor was diagnosed as atypical peripheral adenoma of the lung.

DISCUSSION

The tumor in the present case was entirely unusual as bronchial adenoma
with its atypical cell configurations. The authors could find only one instance in
the literature, in which the histological structure was similar to this in the photograph
or description of the tumor. The 4th case in the report of Felton et al.\textsuperscript{5} was that
of carcinoid type in a great part and neurilemmoma-like appearance in a small
part. Unfortunately, they did not describe about such unusual structure in detail.

As the other possibility in our case, chemodectoma or carotid body-like tumor
can be assumed on the basis of nesting pattern and of rather bland character of the
tumor cells. Carotid body-like tumor in the lung was first reported in 1958 by
Fig. 6. The tumor cells from prominent nesting pattern or acinar structure.
Fig. 7. There are some lumina, but it is questionable whether they represent true gland structures or not.

Heppelestone, but it is at present a rather uncertain entity. On the other hand, the acinar structure and the association of peculiar lymphocytic infiltration in the reported case suggested a resemblance to 'oat cell carcinoma' or to thymoma. As to such a character, the histological findings of Sobota and Reed's case
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are especially worthy of analysis; the patient had Cushing's syndrome, hypokalemic alkalosis and three pulmonary adenomas, one of which was of carcinoid type and two others suggested a common cell origin with epithelial thymomas and oat cell carcinomas. In our case thymoma-like shadow was found on x-ray film. However, on histomorphological analysis of the tumor, metastasis from a thymic tumor could be ruled out. In any way, we can point out the possibility that this atypical growth develops into the thymoma-like structure and/or into oat cell carcinoma.

On the histogenesis of peripheral adenoma of the lung, a number of investigators have assumed that the tumor originates from the lining epithelium of small bronchi, bronchioli or possibly even from respiratory bronchioli since the tumor in problem may develop as multicentric lesions, and a close interrelationship between tumor cells and the lining epithelium has been noted. We could confirm that the tumor had arisen directly from the lining epithelium and significant relationships between them were indicated at the margin of the tumor. Despite of some controversy about the histogenesis of usual bronchial adenoma, it is well known that in the majority of cases it arises in the mucous glands or their ductal epithelium around major bronchi and it is predominantly composed of carcinoid or cylindroid type as well as the peripherally located adenoma is. In contrast to a higher frequency of carcinoid type adenoma in the Western countries, the incidences of cylindroid and carcinoid types of pulmonary adenomas were almost equal in Japan. Generally speaking, the rate of incidence of carcinoid tumors in Japanese race is significantly lower than those in Anglo-American races. Therefore, such constitutional differences may play some role in the morphological manifestation of peripheral adenoma of the lung.

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References


Fig. 8. In this place the cells are elongated and spindle-shaped, taking a palisaded arrangement. The nuclei with relatively high chromatin contents are not fairly uniform.

Fig. 9. A prominent lymphocytic infiltration is associated in a certain part of the tumor.

Fig. 10. Direct transition from bronchiolar lining epithelium to the tumor cells is clearly recognizable.
22) Kikuchi, A. Adenoma of the trachea and bronchus — statistical studies of 60 patients reported in Japan. *Nippon acta radiologica*, 1965, 24, 1189–1203.