Primary Squamous Cell Carcinoma of the Thyroid with Severe Tracheal Invasion: A Case Report

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A 64-year-old Japanese man visited a hospital with symptoms of cough and wheezing. His cervical and chest computed tomography (CT) revealed a hypodense mass (47 × 52 × 63 mm) occupying from the right thyroid lobe to superior mediastinum, with compression and marked lateral displacement of the trachea to the left as shown in Figure 1a. Thyroid-associated data were almost normal: thyroid stimulating hormone, 0.429 μ IU/mL (reference range, 0.400–6.000 μ IU/mL); free tri-iodothyronine, 2.49 pg/mL (reference range, 2.50–4.20 pg/mL); free thyroxine, 0.90 ng/dL (reference range, 0.80–1.60 ng/dL); TgAb antibody, <10.0 IU/mL (reference range, <10.0 IU/mL); and thyroglobulin, 8.89 ng/mL (reference range, 0.00–33.70 ng/mL). Tumor makers (CEA, CA19–9, PSA, cytokeratin 19 fragment, pro-gastrin-releasing peptide, and calcitonin) were almost within normal range. Bronchoscopy showed severe stricture of the trachea and invasion of tumor as shown in Figure 1b. In addition, histological examination of tracheal biopsy showed a highly malignant epithelial proliferation with diffuse and infiltrative features as shown in Figure 1c. Atypical cells had anisokaryosis and atypical mitoses, and some tumor cell areas presented keratinization and necrotic foci. Immunohistochemical staining showed that p40 was positive and p16 was partially positive. CD5 and C-kit were negative, both of which are usually positive for mediastinal tumor (data not shown).

Primary squamous cell carcinoma of the thyroid (PSCCT) is a very rare thyroid malignancy, which accounts for less than 1% of all thyroid malignancies [1, 2]. Most patients with PSCCT have an enlarging neck mass together with dysphonia, dysphasia and/or dyspnea, although such symptoms are not obvious until the tumor becomes pretty large [3]. However, our patient did not notice his tumor by himself, although his thyroid mass became large and marked lateral displacement of the trachea to the left. In general, the invasion of thyroid cancer to the trachea is rare, but if it occurs, airway bleeding and obstruction can lead to death [4]. Most important point of this case is that subjective symptoms with PSCCT are sometimes very poor until the tumor is enlarged.

PSCCT is usually highly aggressive; its prognosis is very poor compared to other thyroid carcinomas, and a median survival time in subjects with PSCCT is as short as about 9–12 months [1], depending on the extent of the tumor resection and adjuvant radiotherapy and/or chemotherapy [5]. There is no consensus for PSCCT management and therapy because of the rarity of cases with PSCCT. Thus, complete removal of the tumor with operation has an advantage for survival, while it is considered PSCCT shows relative resistance to radiotherapy and poor response to chemotherapy [1]. Our patient's PSCCT was diagnosed as squamous cell carcinoma based on his-

Fig. 1. **a** Cervical and chest computed tomography revealed a hypodense mass ($47 \times 5 \times 2 \times 63$ mm) spreading from the right thyroid lobe to superior mediastinum. This tumor was accompanied by compression of the trachea and marked lateral displacement of the trachea to the left. **b** Bronchoscopy showed severe stricture of the trachea and invasion of tumor. **c** HE staining showed a highly malignant epithelial proliferation with diffuse and infiltrative features. Atypical cells had anisokaryosis and atypical mitoses, and some tumor cell areas presented keratinization and necrotic foci.

tological findings. In addition, his PSCCT was complicated with lung metastasis. Therefore, we performed radiation therapy (a total of 59.4 Gy) and chemotherapy (a total of 14 courses of cisplatin and docetaxel combination therapy), which is standard therapy for squamous cell carcinoma. This therapy was pretty effective for our patient, although in general PSCCT is a highly aggressive malignancy with poor prognosis. As a result, our patient survived for more than 12 months after its diagnosis.

Statement of Ethics

The patient has given written informed consent to publish this case including the publication of images.

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Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Y.I. and T.A. researched data and wrote the manuscript. K.K., Y.O., K.N., and Y.M. researched data and contributed to the discussion. H.K., N.O., and K.T. reviewed the manuscript.