Clinical case

Pulmonary leiomyosarcoma metastatic to the thyroid gland:
Case report and review of the literature

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Abstract

Background. – Leiomyosarcoma metastatic to the thyroid is extremely rare. Especially, metastasis of pulmonary leiomyosarcoma to the thyroid is extremely rare—only one such case has been previously reported. Case presentation. – A 55-year-old woman presented with a chief complaint of a cough of 1.5 months duration. Chest radiography (PA view) and chest computed tomography revealed 1 cm sized subpleural nodule in left apical lung and a 8.3 × 4 cm sized, lobulated mass in anterior segment of left upper lobe of the lung. We decided on left upper lobectomy and excision by video-assisted thoracoscopic surgery (VATS). They were leiomyosarcomas. During follow-up chest computed tomography at 23 months after first surgery, we noticed that a nodule on the left lobe of the thyroid gland had increased in size over 3 months. The patient underwent total thyroidectomy and central lymph node dissection. Immunohistochemical staining showed that tumor cells were positive for smooth muscle actin, focal positive for desmin and positive for vimentin, but negative for CD34 and S-100 protein. C-kit staining showed focal, weak positivity. The Ki-67 proliferation index was around 30–40%. Conclusions. – Our case represents the first report of pulmonary leiomyosarcoma metastatic to the thyroid, although extrapulmonary leiomyosarcomas metastatic to the thyroid is encountered infrequently.

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1. Introduction

Leiomyosarcoma metastatic to the thyroid is extremely rare. Although the thyroid gland has a rich blood supply, the incidence of metastases to the thyroid gland is only 2% [1]. However, malignancies to the thyroid gland are not infrequent at autopsy, and are encountered in 2% to 24% of patients with a malignancy [2,3]. However, metastasis of pulmonary leiomyosarcoma to the thyroid is extremely rare—only one such case has been previously reported. Here, the authors present a case of pulmonary leiomyosarcoma metastatic to the thyroid gland.

2. Case report

A 55-year-old woman presented with a chief complaint of a cough of 1.5 months duration. Chest radiography (PA view)
and chest computed tomography revealed a 1 cm sized subpleural nodule in left apical lung and an 8.3 × 4 cm sized, lobulated mass in anterior segment of left upper lobe of the lung, abutting the chest wall near the anterior arc of the 4th to 6th ribs without adjacent rib destruction or atelectasis in the adjacent lung. There was no evidence of airway obstruction or regional lymphadenopathy, and no malignant cells were observed in pleural and bronchial washing fluid. The masses were suspected to be a spindle cell tumor by core biopsy. We decided on left upper lobectomy and excision by video-assisted thoracoscopic surgery (VATS). On gross examination, they measured 9 × 5 × 4 cm and 2.5 × 1.8 × 0.3 cm, respectively and grossly were well-demarcated, white to yellow-coloured, lobulated with geographic necrosis.

They both abutted the pleural surface and adjacent bronchus and revealed they were leiomyosarcomas with an average of 13 mitotic figures per 10 high-power fields on microscopic examination. Both tumors showed necrosis and invasion of visceral pleura, but no regional lymph node metastasis was found. The tumor cells were spindle-shaped and cystiform. Hematoxylin-eosin staining revealed a fascicular pattern of spindle-shaped cells with increased cellularity, and abundant eosinophilic cytoplasm with hyperchromatic, bizarre, malformed, and megakaryocytic blunt-ended nuclei. Immunohistochemical staining showed that tumor cells were strongly and diffuse positive for smooth muscle actin and less strongly, focal positive for desmin. In addition, they were also diffusely positive for vimentin, but negative for CD34 and S-100 protein. C-kit staining showed focal, weak positivity. The Ki-67 proliferation index was around 30–40%. Chemotherapy (doxorubicin, 60 mg/m², 1 day per 3 weeks for three cycles) was given after surgery. During follow-up chest-computed tomography at 23 months after first surgery, we noticed that a nodule on the left lobe of the thyroid gland had increased in size over 3 months. Thyroid hormone assay results were within normal limits, except for an elevated thyroid stimulating hormone level (7.79 μIU/ml). The patient underwent total thyroidectomy and central lymph node dissection at 26 months after first surgery. On gross examination, the nodule had an enlarged round appearance, and in section, the cut-surface showed a well-demarcated, firm, whitish, oval mass that was not encapsulated and invading into and effaced the thyroid parenchyma. Under the microscope, its cellular appearance resembled that of the primary tumors (Fig. 1). In addition, tumor cells extended beyond the tumor and thyroid capsules, and vascular invasion was also evident. Immunohistochemical staining findings were identical to those of the primary pulmonary leiomyosarcomas (Figs. 2, 3). The patient has since been followed for 4 months after thyroidectomy with evidence of iliac bone metastasis by positron emission tomography/computed tomography.

3. Discussion

Although tumors of the thyroid are usually primary, metastasis must be considered in patients with a history of cancer. However, metastatic malignant tumors of the thyroid are extremely rare. Metastatic thyroid tumors represent between...
1.4% and 2.5% of all thyroid cancers [1] and between 0.05% and 0.1% of thyroid diseases, [4] but incidences at autopsy are higher. Autopsy series have demonstrated metastatic disease incidences in the thyroid of 1.25% to 24.4% among patients that succumb to primary or metastatic cancer [2,3,5]. The most frequently reported primary cancers are clear cell renal carcinoma, bronchial carcinoma, and breast cancer [6,7]. The clinical presentations of metastatic thyroid tumors may be similar to those of primary thyroid tumors, and include neck enlargement, hoarseness, stridor, dysphagia, and odynophagia.

Pulmonary leiomyosarcoma may originate either in the interstitium of alveolar walls or from primitive mesenchymal cells. Parenchymal leiomyosarcoma grows in the interstitium and produces a rounded well-defined and demarcated lung lesion resembling any peripheral benign, malignant or solitary metastatic lesion. In the described case, sharply defined and demarcated tumor margins from surrounding lung, the absence of lymph node metastasis, and the low attenuation value of the mass on computed tomography scans did not favor bronchogenic carcinoma [8–11]. Pulmonary leiomyosarcomas do not have any characteristic clinical or radiological findings and are diagnosed using histopathological findings. The microscopic pathological features of leiomyosarcoma may be confused with the more common spindle cell carcinoma, several squamous cell carcinomas, benign leiomyoma, solitary metastasis of a melanoma, and metastatic leiomyosarcoma [12]. Using a MEDLINE search (1971–2010) for the terms, “pulmonary leiomyosarcoma”, “metastatic thyroid tumors”, and “metastatic leiomyosarcoma”, we surveyed the literature for previous cases of pulmonary leiomyosarcoma metastatic to the thyroid. Only one such previous case reported by Shimota et al. in 1991 was identified [13]. The authors described the autopsy findings of leiomyosarcoma originating in the right lung in an 86-year-old man. The solid tumor measured 6 × 7 × 7 cm, and its cut-surface appeared grayish to yellowish and contained areas of hemorrhage and necrosis. The lungs contained scattered nodular lesions of less than 2 cm in diameter. Metastatic lesions were also confirmed in other tissues, including the heart, pericardium, liver, spleen, left adrenal gland, and thyroid. Microscopically, 14 mitoses were detected per 10 high-power fields. An immunohistochemical study revealed positivity for smooth muscle myosin and actin and negativity for CEA, which in combination, appeared to be important for differential diagnosis and the determination of histogenetic type. In conclusion, our case represents the first report of pulmonary leiomyosarcoma metastatic to the thyroid, although extrapulmonary leiomyosarcomas metastatic to the thyroid is encountered infrequently. No definitive treatment is available for this type of metastasis, but total thyroidectomy followed by external beam radiation therapy with or without chemotherapy may prolong survival [14].

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References