

Malignant glomus tumor of the thyroid gland: a case report

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Abstract

Objective: Primary malignant glomus tumors of the thyroid gland are a rare occurrence.

Methods: A 68-year-old man found a mass on the right side of his neck in October 2017. An X-ray examination on 9 January 2018 showed multiple round reinforced masses in both sides of the lung. Computed tomography imaging of the neck showed a low-density mass on the right side of the thyroid gland.

Results: The immunohistochemistry results were positive for smooth muscle actin, calponin, collagen IV, and Ki-67 60%. The patient received chemotherapy starting on 17 January 2018, 8 February 2018, and 11 March 2018. The chemotherapy drugs included ifosfamide, epirubicin, and cisplatin. However, the patient subsequently developed multiple organ failure and died in April 2018, approximately 6 months after the initial discovery of the mass in his neck.

Conclusions: Primary malignant glomus tumors of the thyroid gland are rare, and examination of their pathology and immunohistochemistry is vital for making an accurate final diagnosis. This case also indicates that primary malignant glomus tumors of the thyroid gland may have a poor prognosis, despite chemotherapy.

Keywords

Malignant glomus tumor, thyroid, microscopy, immunohistochemistry, prognosis, chemotherapy

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Introduction

Primary malignant glomus tumors of the thyroid gland are rare and may be associated with a poor prognosis and distant organ metastasis. To the best of our knowledge, only one case of a primary malignant glomus tumor in the thyroid gland has been reported in the English literature.¹ Here, we report another patient with such a tumor who only survived for about 6 months after the initial incidental detection of a thyroid mass.

Case report

Written informed consent was obtained from the patient for the treatments and his relatives agreed to the publication of this case report. A 68-year-old man found a mass on the right side of his neck in October 2017. At that time, the mass was approximately $13 \times 15 \times 5$ mm but was painless, and the patient therefore largely ignored it. However, the mass gradually

increased in size and became painful, with paroxysmal swelling pain radiating to the top of the head. A biopsy was performed at the outpatient department on January 9, 2018, but the doctor in the Pathology Department was unable to make a certain diagnosis at that time. However, an X-ray examination conducted on the same day showed multiple roundish reinforced masses in the bilateral lungs (Figure 1), while neck computed tomography showed a low-density mass at the right side of the thyroid gland (Figure 1).

The pathology report on 12 January 2018, indicated a soft tissue malignant tumor, including regular, circular tumor cells with clear boundaries (Figure 2). The tumor cells were moderately heterotypic, and the cytoplasm showed transparent faint staining with hematoxylin and eosin. Tumor necrosis was detected, with a mitotic count $> 5/50$ high-power fields (HPF). Immunohistochemistry (Figure 3) analysis revealed the tumor to be positive for

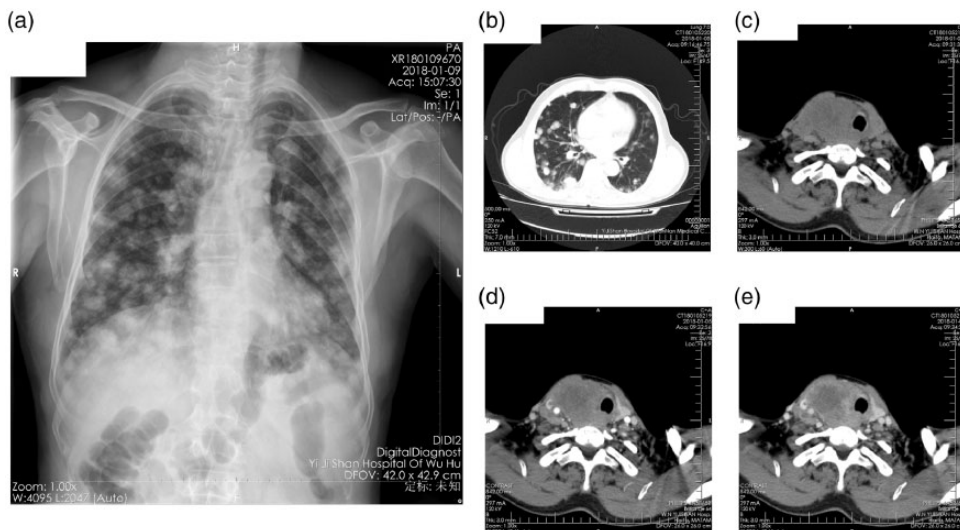


Figure 1. (a) X-ray examination showing right-sided thyroid mass and metastatic tumor in both sides of the lung (red arrow). (b) Lung computed tomography (CT) showing metastatic tumor in both sides of the lung (red arrow). Neck CT showing (c) right-sided thyroid mass (red arrow), (d) right-sided thyroid mass in arterial phase (red arrow), and (e) right-sided thyroid mass in venous phase (red arrow).

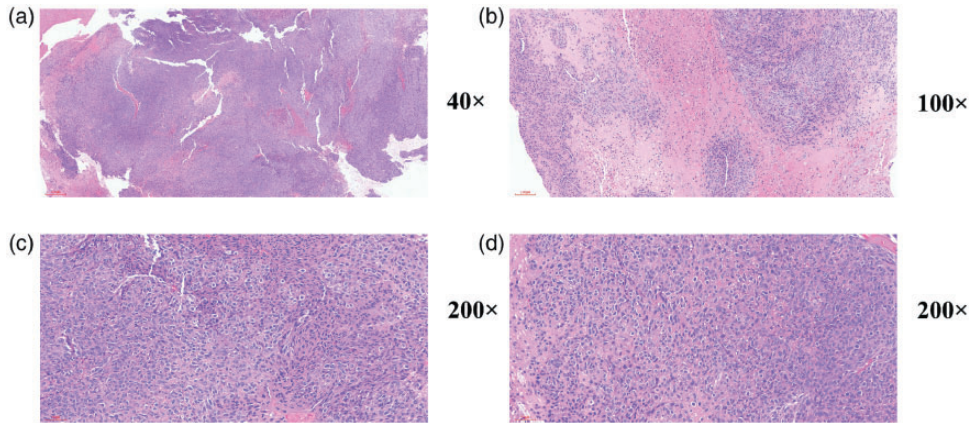


Figure 2. (a, b) Tumor-intervening capillaries were thin and abundant, including sinusoids, showing marked hemorrhage and necrosis (hematoxylin and eosin; HE $\times 40$). (c, d) Tumor cells were circular with clear boundaries, with faint, transparent cytoplasmic staining with HE. Tumor cells showed moderate heterotopia, included vacuoles and small central nucleoli, and demonstrated mitotic activity (HE $\times 200$).

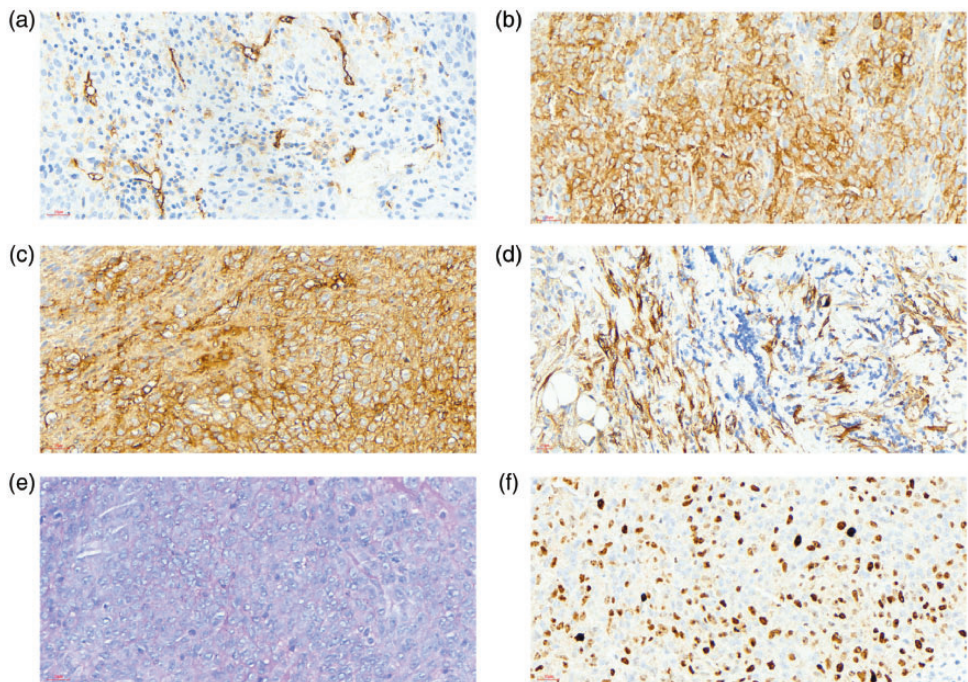


Figure 3. (a) Intervening capillaries stained with CD31 were thin and abundant by immunohistochemistry (IHC; $\times 400$). (b) Tumor cells were immunopositive for SMA, (c) collagen type IV, and (d) calponin (all IHC $\times 400$). (e) PAS staining shows clear tumor cells. (f) Tumor cells showed increased proliferation as demonstrated by increased Ki67 staining (IHC $\times 400$).

smooth muscle actin (SMA), calponin, collagen IV, periodic acid-Schiff (PAS), and Ki67:60%, and negative for cytokeratin AE1/AE3, epithelial membrane antigen (EMA), thyroglobulin, calcitonin, cytokeratin (CK)19, CK7, p63, calretinin, cytokeratin CAM5.2, desmin, S-100, CD99, CD34, CD31, CD21, CD23, CD56, synaptophysin (Syn), and chromogranin A (CgA). Neck ultrasound examination on 7 March 2018, showed an 80 × 44 × 59 mm solid occupying mass with clear margins and heterogeneous internal hypoechoicity in the right side of the neck, invading the right lobe of the thyroid gland, thyroid gorge, and part of the left side of the thyroid gland. Signals indicated a rich tumor blood flow. The mass was diagnosed as a malignant glomus tumor of the thyroid gland. The patient received chemotherapy starting on 17 January 2018, 8 February 2018, and 11 March 2018, including ifosfamide, epirubicin, and cisplatin. However, despite chemotherapy, a metastatic pulmonary mass developed about 6 months after the initial detection of the mass, followed by multiple organ failure, and the patient died within 4 months of diagnosis.

Discussion

We searched the PubMed database from January 1975 to May 2018 using the keywords “malignant glomus tumor” and “thyroid gland”, but only identified one reported case published in English.¹ Chung et al.¹ reported a patient with a malignant glomus tumor in the thyroid gland who died from multiple metastases in his brain and lung about 2.5 years after detection. In the present case, the patient only survived for about 6 months after the incidental detection of a thyroid mass. These cases suggest that this disease may be associated with a poor prognosis, including distant organ metastasis, especially in the lung, after a relatively short duration.

Malignant glomus tumors are usually located in the kidney² and foot,³ but may sometimes occur at other sites in the gastrointestinal tract⁴ or respiratory tract.⁵ However, malignant glomus tumors occurring in the thyroid gland are rare.¹

Folpe et al.⁵ proposed that malignant glomus tumors should meet the following diagnostic criteria: tumors located deep in the body, at least 2 cm in size, with atypical mitotic figures in >5/50 HPF, with moderate to high nuclear grade. The current tumor was 80 × 44 × 59 mm located in the right lobe of the thyroid gland, the cells showed moderate heterotypia, and the mitotic count was >5/50 HPF.

Malignant glomus tumors in the thyroid region need to be differentiated from various other tumors, including medullary carcinomas, undifferentiated carcinomas, leiomyosarcomas, hemangiosarcomas, synovial sarcomas, and dendritic sarcomas. Medullary carcinomas include abundant blood vessels or sinuses in the stroma, amyloid deposits, or positive immunohistochemical neuroendocrine markers, such as calcitonin, which contribute to this diagnosis. However, the immunohistochemistry results in the present patient were negative for calcitonin, CD56, Syn, and CgA, thus excluding a diagnosis of medullary carcinoma. Undifferentiated carcinomas are composed of epithelial carcinoid cells and spindle sarcomatoid cells, with an immunohistochemistry profile positive for CK19, CK7, and EMA. However, these three markers were negative in the current patient, thus ruling out the possibility of undifferentiated carcinoma. Leiomyosarcomas show distinct atypia with a bundle arrangement, and spindle-shaped cells with blunt rounded ends. Leiomyosarcomas are positive for desmin, while the present case was negative, thus excluding a leiomyosarcoma. Hemangiosarcomas include anastomotic fissures with red blood cells and large, heterogeneous endothelial cells in the fissures.

Immunohistochemically, hemangiosarcomas are positive for CD34 and CD31, while the current patient was negative for both these antigens. Synovial sarcomas show bidirectional differentiation and are positive for CAM5.2 and CD99, while the present patient's tumor was negative for both CAM5.2 and CD99, which excluded a diagnosis of synovial sarcoma. Tumor cells are arranged in coils with lymphocyte infiltration in dendritic cell sarcomas, which are also immunohistochemically positive for CD21 and CD23. The immunohistochemistry results in the present patient were negative for CD21 and CD23, which ruled out a dendritic cell sarcoma.

The treatment options for malignant glomus tumors without a head and neck origin include complete surgical removal, radiotherapy, and chemotherapy.¹⁻⁵ One patient with a malignant glomus tumor of the kidney² received palliative radiation therapy and one cycle of gemcitabine, docetaxel, doxorubicin, and dacarbazine. However, the radiation therapy failed to improve the patient's pain. Chung et al.¹ reported the first case of a malignant glomus tumor in the thyroid gland, measuring 3.6 × 3.5 × 2.8 cm, located in the right thyroid gland lobe, with occasional cellular atypia and necrosis, mitotic activity up to 7/10 HPF, with moderate to severe cellular pleomorphism. This previous patient underwent complete thyroidectomy and chemotherapy with adriamycin/CDDP, but subsequently developed metastatic cerebral and pulmonary masses within 30 months. Similarly, the present case developed pulmonary metastasis and died within a short space of time, despite receiving chemotherapy.

In conclusion, primary malignant glomus tumors in the thyroid gland are rare, and pathological and immunohistochemical examinations are required to

make a final accurate diagnosis. In addition, primary malignant glomus tumors in the thyroid gland may have a poor prognosis despite chemotherapy.

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Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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