Unusual Granular Cell Tumor of the Trachea Coexisting With Papillary Thyroid Carcinoma and Masquerading as Tracheal Invasion of Recurred Thyroid Carcinoma

A Case Report

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Abstract: This was an extremely rare case of unusual granular cell tumor of the trachea coexisting with recurrent papillary thyroid carcinoma. We initially misdiagnosed this patient as having tracheal invasion from recurrent thyroid carcinoma even after a computed tomography scan and fine-needle aspiration cytology. Clinicians should be aware of the possibility of granular cell tumor of the trachea occurring simultaneously with papillary thyroid carcinoma.

KEY FINDINGS
- This was the second reported case of unusual granular cell tumor of the trachea coexisting with papillary thyroid carcinoma.
- Clinicians should be aware of the possibility of granular cell tumor of the trachea occurring simultaneously with papillary thyroid carcinoma.

INTRODUCTION
Granular cell tumor is a rare, typically benign neoplasm usually of Schwann cell origin. Granular cell tumor can occur anywhere in the body. However, majority of granular cell tumors involve the head and neck region, and they have only rarely been reported to involve the trachea. Because of its rarity and unusual location, granular cell tumor of the trachea may be clinically misdiagnosed as thyroid mass or tracheal invasion from thyroid carcinoma. Herein, we present a case of unusual granular cell tumor of the trachea coexisting with recurrent papillary thyroid carcinoma and masquerading as tracheal invasion of recurrent thyroid carcinoma.

CASE REPORT
A 45-year-old female presented to our clinic with blood-tinged sputum, cough, and shortness of breath of 3 weeks duration. The patient had a history of left thyroid lobectomy due to thyroid carcinoma at another hospital, and she was lost to follow-up after surgery. The laboratory test and chest radiography findings were normal. Rigid bronchoscopy revealed a smooth, polypoid, yellow lesion emanating from the anterior cervical tracheal wall and occluding >90% of the airway (Figure 1). Computed tomography (CT) scan revealed a suspicious recurrent thyroid carcinoma with invasion into the tracheal lumen and left lateral neck metastasis (Figure 1). Fine-needle aspiration cytology was performed under ultrasound guidance, and the left neck lymph node was interpreted as being consistent with metastatic papillary carcinoma.

Based on these observations, the patient was initially diagnosed with recurrent papillary thyroid carcinoma with tracheal invasion and left lateral neck metastasis. Initially, we performed tracheal mass removal by rigid bronchoscopy with electrocautery and argon plasma coagulation. After airway stabilization, completion thyroidectomy and left modified radical neck dissection were performed. During surgery, thyroid lobe and trachea were easily dissected, and there was no evidence of neoplastic infiltration into tracheal rings. Therefore, we completed the operation without tracheal ring resection. Pathological examination of the thyroid and left neck lymph nodes revealed recurrent and metastatic papillary thyroid carcinoma. However, pathological diagnosis of the tracheal mass was consistent with granular cell tumor (Figure 2). The tracheal mass was strongly positive for S-100 protein on immunohistochemical staining (Figure 3). This case was finally diagnosed as unusual granular cell tumor of the trachea coexisting with recurrent papillary thyroid carcinoma. The postoperative course was uneventful. At the 13-month follow-up, the patient was asymptomatic and there was no evidence of local recurrence or metastasis.

DISCUSSION
Granular cell tumor occurs in a wide variety of organs, with the most common involvement site being the head and neck region. The reported rate of respiratory tract involvement is very low. In conclusion, clinicians should be aware of the possibility of granular cell tumor of the trachea occurring simultaneously with papillary thyroid carcinoma and masquerading as tracheal invasion of recurrent thyroid carcinoma.
involvement is ~11%, and the laryngobronchial tree is involved more often than the trachea.1–5 Therefore, if an extremely rare granular cell tumor occurs in the trachea, this lesion may be clinically misdiagnosed as tracheal invasion from thyroid carcinoma.3 The misdiagnosis of granular cell tumor of the trachea may be occurred in patients with thyroid nodules or previous history of operation for thyroid carcinoma, as in this patient.

Granular cell tumor of the trachea is asymptomatic.4,5 In some cases, patients may present with respiratory symptoms, such as progressive dyspnea unresponsive to bronchodilator and steroid therapy, cough, wheezing, and hemoptysis.1,2,4,5 A careful history, physical examination, radiologic examination, and a high index of suspicion are necessary for making the diagnosis of granular cell tumor of the trachea.1,2 Laryngoscopy and bronchoscopy can reveal the extent of the lesion and a biopsy can be performed.4 CT or magnetic resonance imaging (MRI) may be helpful to evaluate the intraluminal extent of the lesion, and extraluminal spread to the esophagus, mediastinum, or trachea.1,2 The diagnosis of granular cell tumor should be confirmed by histopathologic examination with immunohistochemical staining such as S-100 protein and neuron-specific enolase.4 The differential diagnosis for granular cell tumor of the trachea includes all subglottic masses, such as benign or malignant tumors of the trachea, esophagus, thyroid, and mediastinum.1,2,4

The definitive treatment of granular cell tumor of the trachea is complete surgical excision including bronchoscopic excision and tracheal resection.1–5 Tracheal resection has the best cure rate; however, it also has a significantly high mortality rate.2 In this patient, we initially planned to perform tracheal resection. However, there was no evidence of invasion from both granular cell tumor of the trachea and recurrent papillary carcinoma. Therefore, in this patient, granular cell tumor was treated by bronchoscopic excision without tracheal resection.

There were some unique characteristics of this case. First, this was the second reported case of unusual granular cell tumor coexisting with papillary thyroid carcinoma.3 Second, we initially misdiagnosed this patient as having tracheal invasion from recurrent thyroid carcinoma even after a CT scan and fine-needle aspiration cytology. Misdiagnosis occurred as the

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FIGURE 1. (A) Rigid bronchoscopy reveals a smooth, polypoid, yellow lesion emanating from the anterior cervical tracheal wall and occluding >90% of the airway. (B) Computed tomography scan of the neck reveals a 2.9 × 2 cm enhancing soft tissue mass in the visceral space, with invasion into the tracheal lumen.

FIGURE 2. Pathological examination shows that tumor nests are composed of large round cells with eosinophilic and granular cytoplasm (hematoxylin and eosin stain, ×200).

FIGURE 3. The tracheal mass is strongly positive for S-100 protein on immunohistochemical staining (S-100, ×200).
patient had a previous history of operation for thyroid carcinoma and a suspicious recurrent thyroid carcinoma with invasion into the tracheal lumen and left lateral neck metastasis based on the interpretation of CT scans. Therefore, clinicians should be aware of the possibility of granular cell tumor of the trachea occurring simultaneously with papillary thyroid carcinoma.

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