Case Report

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Adenomatous hyperplastic intratracheal ectopic thyroid tissue: a case report

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Abstract

Intratracheal ectopic thyroid (ITET) is a rare disease, with limited cases reported in the literature. ITET is an unusual congenital abnormality and can be easily mistaken for a respiratory illness. We present a case of a 61-year-old man with a history of slight discontinuous hemoptysis for 2 years. A tracheal mass, which appeared to be connected to the left thyroid gland, was found by chest computed tomography scan. Ultrasound revealed one suspiciously malignant, solid and hypoechoic nodule in the left thyroid gland. After the thyroid origin of the mass was confirmed by bronchoscopic biopsy, the patient underwent segmental resection and anastomosis of the trachea, together with left thyroidectomy. Histopathology of the tracheal tumor showed adenomatous hyperplastic ITET, and the orthotopic left thyroid gland showed nodular goiter with atypical adenomatous hyperplasia. Clinical suspicion is warranted in patients presenting with a tracheal tumor seemingly connected to the thyroid gland, particularly in patients who have imaging features suggestive of a malignant tumor in the orthotopic thyroid but without confirmative histopathology of malignancy before surgery.

Keywords

Intratracheal ectopic thyroid, ectopic, thyroid, diagnosis, treatment, adenomatous hyperplasia

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Introduction

Ectopic thyroid is a rare disease that refers to thyroid tissue lying in locations other than the normal neck region. Ectopic thyroid is believed to be caused by failure of migration of the original thyroid tissue in the early stage of embryogenesis.[1] The mechanism of ectopic thyroid is not clear but it might be related to mutations in the transcription factors thyroid transcription factor 1 (TTF1), forkhead box E1 (FOXE1), and paired box 8 (PAX8).[2] Ectopic thyroid tissue may be located anywhere along its embryologic migration path, including the base of the tongue, lateral neck, carotid artery, lung tissue, intestine, gallbladder, and kidney.[3] Compared with other types of ectopic thyroid, intratracheal ectopic thyroid (ITET) is a rarer congenital abnormality, and its clinical symptoms can be easily confused with respiratory illness, which can lead to misdiagnosis and affect the treatment strategy. To remind clinicians of this disease, we present here a case of adenomatous hyperplastic ITET.

Case report

A 61-year-old man was admitted to a local hospital for backache. Computed tomography (CT) scan of the chest showed a large mass arising from the left postero-lateral wall of the trachea, with an uncertain connection to the left thyroid gland (Figure 1a-c). The patient was transferred to our hospital for further evaluation. The patient

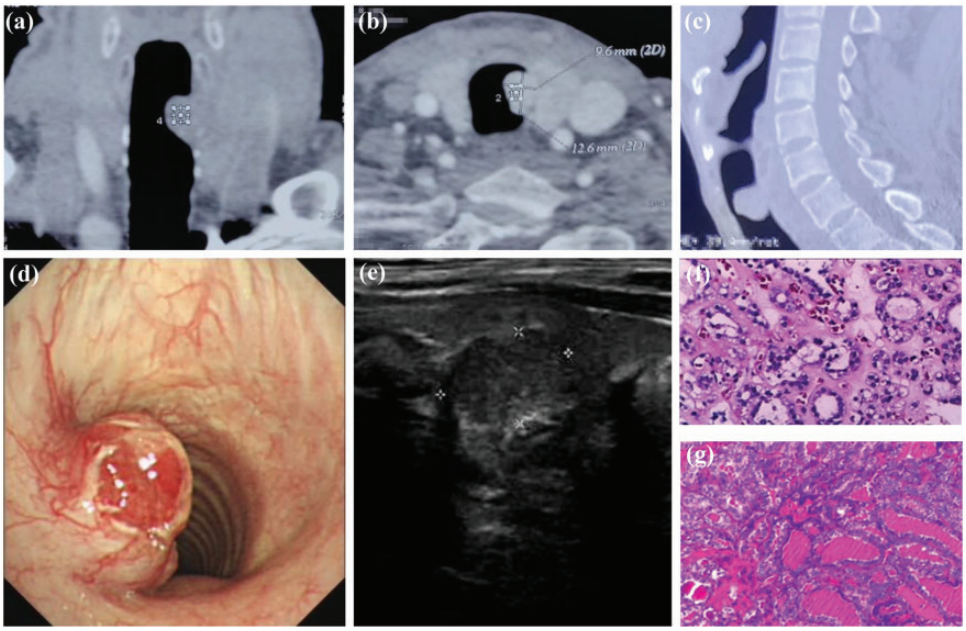

Figure 1. Coronal view (a), axial view (b), and sagittal view (c) in computed tomography scan of the neck without contrast, showing an intratracheal mass immediately below the true vocal folds, with an obscured tracheal wall. Fiberoptic bronchoscopy (d) revealed a submucosal mass on the left tracheal wall occupying approximately 30% of the airway. Thyroid ultrasound (e) revealed one solid and hypoechoic nodule of 14×9 mm in the left thyroid gland, with a hyperechoic area and irregular shape. Fiberoptic bronchoscopic biopsy (f) revealed that the tracheal tumor consisted of mature thyroid tissue (hematoxylin and eosin [H&E] stain, 100× magnification). Histopathologic findings (g) showed hyperplasic thyroid follicular cells containing colloid material (H&E stain, 100× magnification).

reported slight discontinuous hemoptysis for 2 years but no other symptoms associated with respiration. He had no significant past medical history and no family history of thyroid diseases, congenital anomalies, or consanguineous marriage.

When the patient was seen for preoperative laboratory investigation, thyroid function was normal, with a thyrotropin (thyroid-stimulating hormone, TSH) level of 3.67 μIU/mL (normal: 0.35–4.94), a free triiodothyronine (FT3) level of 3.11 pg/mL (normal: 1.71–3.71), and a free thyroxine (FT4) level of 0.92 ng/dL (normal: 0.70–1.48). His thyroglobulin level was 10.94 ng/mL (normal: 3.5–77.0), thyroglobulin antibody (TgAb) level was 62.78 IU/mL (normal: <115), and thyroid peroxidase antibody (TpoAb) level was 35.72 IU/mL (normal: <34).

Preoperative fiberoptic bronchoscopy demonstrated a broad-based mass, with abundant blood supply, originating on the left tracheal wall inferior to the left vocal cord and occupying approximately 30% of the airway (Figure 1d). Thyroid ultrasound revealed several solid and hypoechoic nodules in the left thyroid gland, without obvious enlarged cervical lymph nodes. One of the nodules measured 14×9 mm and had hyperechoic areas and an irregular shape (Figure 1e); it was recorded as Thyroid Imaging Reporting and Data System (TIRADS) 4b by an experienced ultrasound doctor, indicating a >10% risk of malignancy. The pulmonary function test was normal. The tracheal mass biopsy showed thyroid tissue (Figure 1f).

The patient refused a fine-needle aspiration biopsy of the thyroid, so it was difficult to exclude thyroid cancer with invasion of the trachea. For definitive diagnosis and follow-up treatment, segmental resection and anastomosis of the trachea and left thyroidectomy were recommended to the patient. After being informed of the advantages and risks, the patient consented to surgery and signed the informed consent form for complete removal of the tracheal mass. During surgery, a relatively well circumscribed soft mass was found that was connected to the left thyroid gland. Frozen pathology indicated a benign tumor in the left thyroid gland during surgery, so the right thyroid gland was preserved. Histopathologic examination of the tracheal mass revealed mature thyroid tissue with adenomatous hyperplasia, suggestive of ITET with adenomatous hyperplasia (Figure 1g). The orthotopic left thyroid gland also showed nodular goiter with atypical adenomatous hyperplasia. After surgery, the patient was admitted to the intensive care unit; he remained stable but had slight hoarseness. Fiberoptic bronchoscopy showed reduced mobility of the left vocal cord and a free right vocal cord 6 days after surgery. Six months later, the patient underwent a CT scan that showed no thyroid tissue remaining in the trachea; his voice recovered well.

Discussion

The first case of ITET was reported in 1875 by Ziemssen.[4] In general, ITET is a curable disease but can cause potentially fatal obstructions of the upper respiratory tract. The special anatomical location of ITET means that most of its clinical symptoms are related to respiratory obstruction. A previous study suggested that sudden worsening dyspnea was a common symptom of ITET, which could be related to diet, hormone levels, or prior thyroidectomy.[5] Karakullukcu et al.[6] found that TSH levels might play a vital role in sudden worsening symptoms of ITET and that most patients with ITET remain asymptomatic until their TSH levels increase. In our case, the patient had no dyspnea and his pulmonary function test was normal, which might be related to his normal TSH level and that the mass obstructed less than

one-third of the trachea. Other symptoms of ITET include stridor and cough.[5] In our case, the patient had slight discontinuous hemoptysis, which has not been reported previously. The hemoptysis might be related to the abundant blood supply of the tracheal tumor.

Most cases of ITET are found by chance. Chest CT scan has a high sensitivity for detecting ITET, but it is hard to distinguish ITET from other tracheal-occupying lesions, such as hemangioma. Laryngoscopic or bronchoscopic biopsy is helpful for the diagnosis of ITET. Because of the similar clinical symptoms, ITET can be easily misdiagnosed as a respiratory illness; a common misdiagnosis is asthma. Previous reports indicate that many patients are misdiagnosed for several years as having asthma.[7] Moreover, ITET with adenomatous hyperplasia should be distinguished from thyroid cancer with tracheal invasion. Typically, thyroid cancer with tracheal invasion can be diagnosed by detection of a calcified mass in the thyroid gland by imaging and by obvious cervical lymph node metastasis. However, See et al.[8] reported that it is difficult to exclude thyroid cancer with tracheal invasion when CT shows imaging manifestations suspicious for a solid tumor in the thyroid gland and disappearance of the tracheal wall between the thyroid gland and tracheal tumor. These findings are similar to those in our case. Although bronchoscopic biopsy confirmed that the tracheal tumor was thyroid tissue in our case, we could not exclude thyroid cancer with tracheal invasion because of the uncertain connection between the left thyroid gland and tracheal tumor, as well as the imaging features that were suggestive of a malignant tumor in the left thyroid. Thus, we chose to remove the tumor via segmental resection and anastomosis of the trachea for definitive diagnosis and treatment.

The management of ITET is not clearly established. For ITET patients with obvious respiratory obstruction, open access surgery is the mainstay of treatment. Most ITET patients undergo tracheofissure and tracheal repair to remove the tracheal mass. Endoscopic resection with laser is another approach in the management of ITET. The disadvantage of endoscopic laser resection of the tumor is the possibility of residual thyroid tissue after the procedure. One case report indicated that a patient had residual ectopic thyroid accounting for less than 5% of the tracheal lumen 15 months after endoscopic laser surgery.[9] Additionally, patients need to endure the pain of plugging trials and to maintain tracheal intubation for several days after surgery.[10] In the past two decades, segmental resection and anastomosis of the trachea has been reported in only two other cases of ITET with adenomatous hyperplasia.[11,12] We believe that segmental resection and anastomosis of the trachea is suitable for this type of ITET because it reduces the risk of sudden respiratory obstruction caused by residual adenomatous hyperplasia ectopic thyroid tissue after tracheofissure or endoscopic resection with the laser; further studies are needed to confirm this. For treatment of ITET patients after surgery, Kansal et al.[13] recommended lifelong thyroxine suppression to prevent enlargement of residual ectopic thyroid tissue after surgery, although this approach remains controversial.

In summary, we present a rare case of adenomatous hyperplastic ITET that had clinical features similar to those of thyroid cancer with tracheal invasion and yet had benign histopathological findings. CT scan revealed a tracheal tumor seemingly connected to the left thyroid gland, and biopsy by bronchoscopy confirmed that it consisted of thyroid tissue. Clinicians should maintain suspicion for ITET in patients who show a tracheal tumor apparently connected to the thyroid gland, with imaging features of malignant tumor in the

orthotopic thyroid but without confirmative histopathology of malignancy before surgery.

Ethical approval

This case report was authorized for publication by the review board of our institution. Written consent to publish the case report was obtained from the patient.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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References

1. Ibrahim NA and Fadeyibi IO. Ectopic thyroid: etiology, pathology and management. *Hormones (Athens)* 2011; 10: 261–269.
2. De Felice M and Di Lauro R. Thyroid development and its disorders: genetics and molecular mechanisms. *Endocr Rev* 2004; 25: 722–746.
3. Noussios G, Anagnostis P, Goulis DG, et al. Ectopic thyroid tissue: anatomical, clinical, and surgical implications of a rare entity. *Eur J Endocrinol* 2011; 165: 375–382.
4. Paltauf R. Zur kenntniss der schilddrusentumoren im inern des kehlkopfs und der luftohre. *Beitr Pathol Anat* 2020; 11: 71–89.
5. Byrd MC, Thompson LDR and Wieneke JA. Intratracheal ectopic thyroid tissue: a case report and literature review. *Ear Nose Throat J* 2003; 82: 514–518.
6. Karakullukcu B, Guevenc G, Cansiz H, et al. Intratracheal ectopic thyroid: case report and review. *Ear Nose Throat J* 2007; 86: 406–408, 418.
7. Al-Hajjaj MS. Ectopic intratracheal thyroid presenting as bronchial asthma. *Respiration* 1991; 58: 329–331.
8. See AC, Patel SG, Montgomery PQ, et al. Intralaryngotracheal thyroid–ectopic thyroid or invasive carcinoma? *J Laryngol Otol* 1998; 112: 673–676.
9. Chark DW, Oliaei S and Armstrong WB. Ectopic intratracheal thyroid causing airway obstruction. *Laryngoscope* 2010; 120: S144.
10. Pritchyk KM, Thompson LD and Malekzadeh S. Endoscopic management of intratracheal ectopic thyroid. *Otolaryngol Head Neck Surg* 2004; 130: 630–632.
11. Serraj M, Ouadnouni Y, Lakranbi M, et al. Intratracheal ectopic thyroid tissue. *Ann Thorac Surg* 2013; 95: e13–e14.
12. Sung YM, Lee KS, Han J, et al. Intratracheal ectopic thyroid tissue with adenomatous hyperplasia in a pregnant woman. *AJR Am J Roentgenol* 2008; 190: W161–W163.
13. Kansal P, Sakati N, Rifai A, et al. Lingual thyroid. Diagnosis and treatment. *Arch Intern Med* 1987; 147: 2046–2048.