



Ultrasonographic findings of tracheal adenoid cystic carcinoma with thyroid invasion and mimicking thyroid tumors: a case report

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Background: Tracheal adenoid cystic carcinoma (TACC) is a rare, low-grade malignant tumor. The primary TACC usually metastasizes to the lung and bone, rarely involving the thyroid. Although some previous reports have described the imaging features of TACC with thyroid invasion, the multimodal ultrasound findings of TACC with thyroid invasion and mimicking thyroid tumors have not been reported before.

Case Description: A 69-year-old woman who had been experiencing hoarseness for 2 years and a thyroid nodule for 2 months was presented to our clinic. Conventional ultrasound showed a hypoechoic nodule about 33×25×50 mm in the left lobe and isthmus of the thyroid, adjacent to the trachea and extending to the right lobe. Contrast-enhanced ultrasound (CEUS) showed that the nodule was unevenly enhanced, with iso-enhancement in the periphery and hypo-enhancement in most of the central area. Shear wave elastography showed that the maximum Young's modulus of nodules was 237.5 kPa, the minimum was 0.1 kPa, and the average was 60.5 kPa. Triiodothyronine, thyroxine, thyroid stimulating hormone and calcitonin were within the normal range. The patient underwent radical surgery with an uneventful postoperative recovery. Combined with the intraoperative findings and pathological examination, the diagnosis of TACC with thyroid invasion was made.

Conclusions: This rare case shows that TACC invading the thyroid may be manifested as a thyroid tumor on ultrasound. Preoperative pathological examination and comprehensive imaging examination are of great significance for the clinical management of patients. We also reviewed the literature on the imaging findings and clinical performance for TACC with thyroid invasion.

Keywords: Tracheal adenoid cystic carcinoma (TACC); thyroid; multimodal ultrasound; invasion; case report

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Introduction

Adenoid cystadenocarcinoma (ACC) is one of the most common malignant tumors in salivary glands, which accounts for approximately 1% of head and neck malignant tumors (1). ACC is also found in the breast,

lungs, larynx, and trachea (2-5). Tracheal adenoid cystic carcinoma (TACC) originates in the glands of the tracheal and bronchial walls (6). Primary TACC most frequently metastasizes to the lungs and the bone, and it rarely involves the thyroid (7,8). To the best of our knowledge,

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there have been no previous reports in the literature on multimodal ultrasound findings of TACC invading the thyroid presenting as a thyroid tumor. Herein, we report a case of multimodal ultrasound findings of TACC invading the thyroid presenting as a thyroid tumor to improve the understanding of this disease. We also reviewed the literature on the imaging findings and clinical performance for TACC invading the thyroid. We present this case in accordance with the CARE reporting checklist (available at <https://gs.amegroups.com/article/view/10.21037/gS-23-485/rc>).

Case presentation

A 69-year-old woman presented to the thyroid surgery department of our hospital with a complaint of hoarseness for 2 years and a thyroid nodule found for 2 months. The patient presented to a local hospital 2 years ago with hoarseness, and laryngoscopy revealed vocal cord paralysis, the details of which were unknown. Then the patient was followed up regularly in the local hospital. Two months ago, the patient underwent thyroid ultrasound examination in a local hospital, and a thyroid nodule was found, which was considered to be thyroid cancer. Therefore, the patient came to our hospital for further treatment. The patient had undergone total hysterectomy for uterine fibroids 20 years earlier and had an uneventful recovery. The patient had no history of smoking or alcohol consumption. The patient also had no other personal or family history of acute or chronic disease.

Physical examination revealed a mass with a diameter of about 5.0 cm in the left lobe of the thyroid, which was irregular in shape, smooth in surface, without tenderness, and could move up and down with swallowing. The mass was non-pulsatile, and no murmur was heard. Conventional ultrasound showed a hypoechoic nodule about 33×25×50 mm in the left lobe and isthmus of the thyroid, adjacent to the trachea and the surrounding dorsal membrane, and extending to the right lobe (*Figure 1A*). The boundary of the nodule was unclear, the shape was irregular, and there was micro-calcification inside (*Figure 1A*). There was no hypoechoic halo around the nodule, and there was punctate blood flow inside (*Figure 1B*). Contrast-enhanced ultrasound (CEUS) showed that the nodule was unevenly enhanced, with iso-enhancement in the periphery and hypo-enhancement in most of the central area (*Figure 1C*). Shear wave elastography showed that the maximum Young's modulus of nodules was 237.5 kPa, the minimum was 0.1 kPa, and the average was 60.5 kPa (*Figure 1D*). There were no obvious abnormal enlarged lymph nodes in the bilateral neck. Neck computed tomography (CT) showed a low-density mass in the left lobe of the thyroid. The largest section was about 3.8×3.1 cm, with uneven mild enhancement, extending contralateral through the posterior direction of the trachea, and unclear boundaries with the trachea and esophagus (*Figure 2*). Neck CT showed no other abnormal findings. Bronchoscopy revealed bilateral vocal cord paralysis and a new bulge in the upper tracheal segment. Triiodothyronine, thyroxine, thyroid stimulating hormone and calcitonin were within the normal range. Ultrasound-guided thyroid fine needle aspiration (FNA) was performed for the patient. Cytopathology report showed atypical follicular epithelial cells, and repeat puncture was recommended. The patient then underwent ultrasound-guided core needle biopsy (CNB). Hematoxylin and eosin staining (*Figure 3A*) showed that the tumor cells were arranging in a stratified tubular structure. Immunohistochemistry revealed TG (-), TTF-1 (-), PAX-8 (-), CK7 (+), CD117 (+), SMA (+) (*Figure 3B-3D*). Adenoid cystic carcinoma was considered from pathological findings. The clinical stage of the patient is T4NxM0. The patient underwent radical tracheal tumor resection and total thyroidectomy. Based on the intraoperative findings and postoperative pathological results, it was considered that TACC invaded the thyroid. The patient had an uneventful recovery after surgery. No tumor recurrence was found during the 2-year follow-up. All procedures performed in this study were in accordance with the ethical standards

Highlight box

Key findings

- Tracheal adenoid cystic carcinoma (TACC) with thyroid invasion can mimic thyroid tumors on ultrasonographic images.

What is known and what is new?

- The primary TACC usually metastasizes to the lung and bone, rarely involving the thyroid.
- We report a case of TACC with thyroid invasion mimicking thyroid tumors with multimodal ultrasound findings. This helps sonographers increase their understanding of the disease.

What is the implication, and what should change now?

- TACC can invade the thyroid and appear as thyroid malignant tumors on ultrasound images. We should combine the patient's clinical symptoms, laboratory tests, imaging examinations and other examinations to make a comprehensive diagnosis.

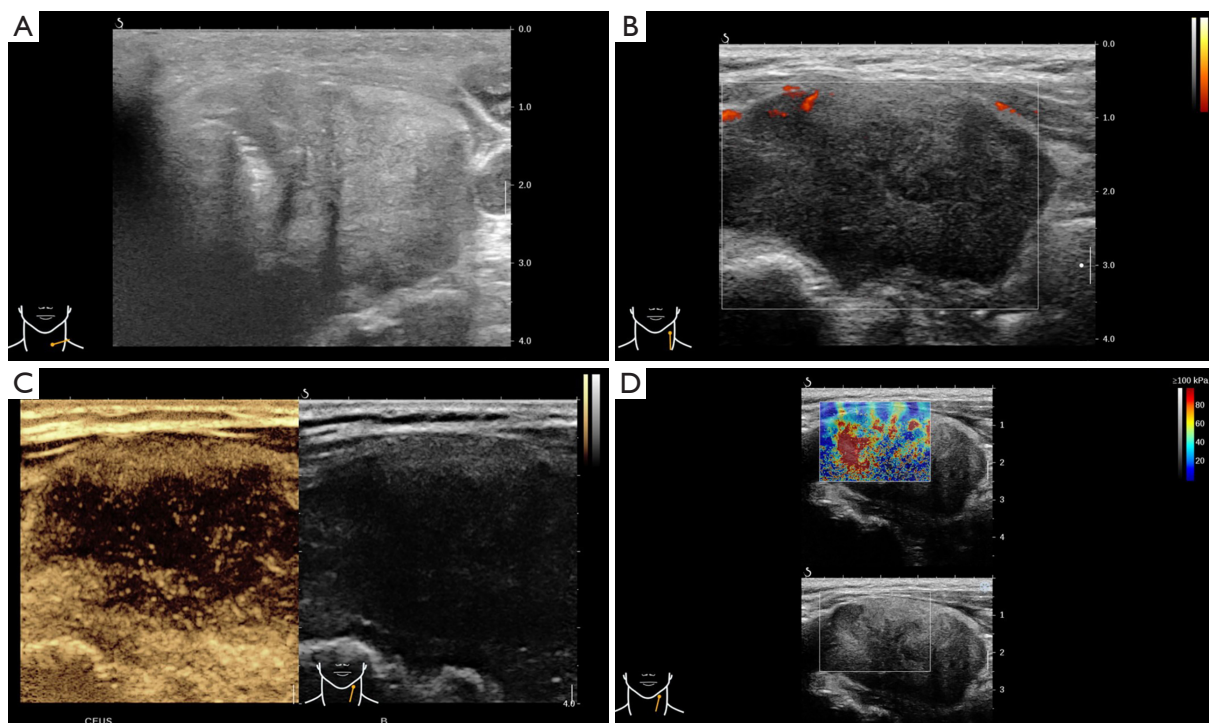


Figure 1 Ultrasound images of tracheal adenoid cystic carcinoma with thyroid invasion. (A,B) Conventional ultrasound shows a hypoechoic nodule in the left lobe and isthmus of the thyroid, adjacent to the trachea and the surrounding dorsal membra. The boundary of the nodule is unclear, the shape is irregular, and there is micro-calcification inside. (C) CEUS shows that the nodule is unevenly enhanced, with iso-enhancement in the periphery and hypo-enhancement in most of the central area. (D) Shear wave elastography shows that the nodule is hard in texture. CEUS, contrast-enhanced ultrasound.

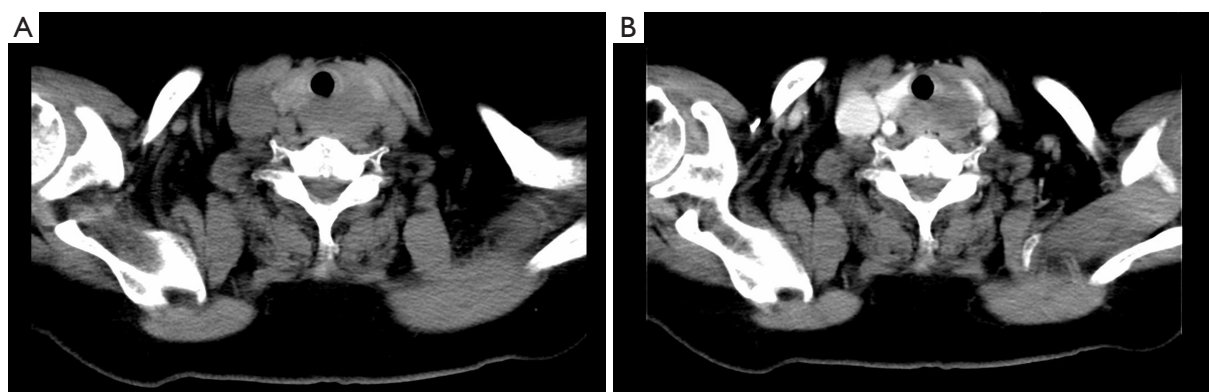


Figure 2 CT images of tracheal adenoid cystic carcinoma with thyroid invasion. (A,B) CT shows a low-density mass in the left lobe of the thyroid, with unclear boundaries with the trachea and esophagus, and heterogeneous mild enhancement. CT, computed tomography.

of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the

editorial office of this journal.

Discussion

Primary tumors of the trachea are rare, accounting for

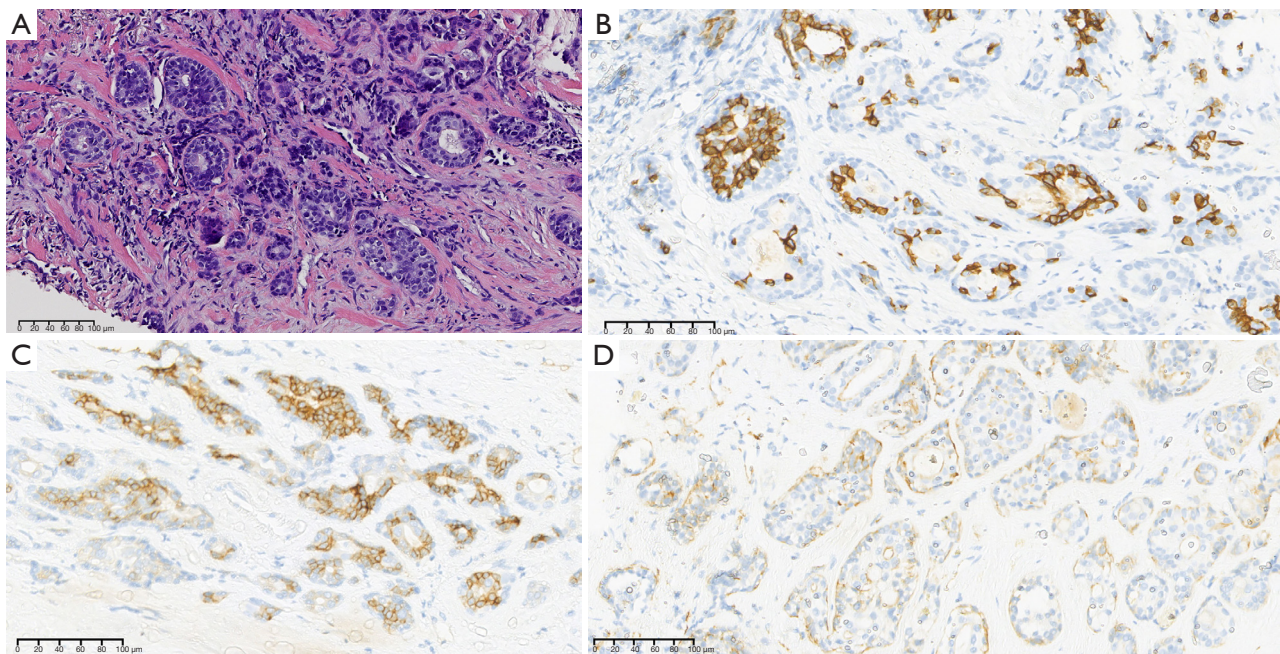


Figure 3 Histopathological images of tracheal adenoid cystic carcinoma with thyroid invasion. (A) Hematoxylin and eosin staining shows that the tumor cells are arranging in a stratified tubular structure. (B-D) Immunohistochemistry shows CK7 (+), CD117 (+), SMA (+).

under 0.1% of the cancer deaths per year, and the main pathological type was squamous cell carcinoma (9,10). The primary TACC usually metastasizes to the lung and bone, rarely involving the thyroid (7,8). Both TACC and granulosa cell tumors can mimic thyroid tumors (11). And there is no literature report on the multimodal ultrasound findings of TACC invading the thyroid as a thyroid tumor.

We reviewed the clinical data and imaging features of twelve patients with TACC thyroid invasion reported in the literature (2,6,12-17). The clinical findings and imaging features of these 12 patients are summarized in *Table 1*. The age of patients ranged from 17 to 77 years old, and there was a significant female predominance, with nine females (75%), three males. The major symptoms found in these patients were dyspnea, hoarseness, cough and breathlessness. The clinical symptoms of the patient we reported were hoarseness, similar to those reported in the literature. Most patients showed an increase in thyroid volume on preoperative imaging examination. The lesions were mainly located in the left lobe, and some were in the right lobe and bilateral lobes. The size of the lesion on convention ultrasound was reported in one case, and the maximum diameter of the lesion was 37 mm. One case reported that the lesion was hypoechoic on conventional ultrasound. In our case, conventional ultrasound showed

that the lesion was hypoechoic, mainly located in the left lobe of the thyroid, and the maximum diameter of the lesion was 50 mm. Eleven patients underwent preoperative CT examination, which showed an enlarged thyroid lobe with contralateral extension and compression of the trachea. Our case showed that the lesion was located in the left lobe and extended to the right lobe, and the boundary with trachea and esophagus was not clear, which was similar to the CT findings of previous cases. Magnetic resonance imaging findings in one patient showed that the tumor was iso- to slight hyperintense relative to muscle on T1-weighted images and of homogeneous high intensity on T2-weighted images. One case showed that free triiodothyronine, free thyroxine, thyroid stimulating hormone, carcinoembryonic antigen, and calcitonin were within normal limits (16). FNA was performed in four patients, two of whom were diagnosed with ACC and two with no definite diagnosis. One patient in the literature review had no definite diagnosis after FNA and was confirmed by CNB, which was similar to the diagnostic process of the patient in our report. Therefore, when FNA is not clearly diagnosed, it may be necessary to perform CNB for patients. CEUS can further guide FNA or CNB, as it can show the blood supply of the lesion and accurately identify the necrotic area. At present, surgery is still the main treatment for ACC with thyroid

Table 1 Tracheal adenoid cystic carcinoma with thyroid invasion in the literature

Author, year	Age (years)	Sex	Symptom	Imaging	Treatment
Zirkin, 1984 (2)	66	F	Enlargement of thyroid nodule and dyspnea	NA	RR
Na, 1995 (12)	33	M	Dyspnea	The encircling mass surrounding the trachea and the circumferential thickening of the tracheal wall were demonstrated in all cases on CT. There was no calcification, no cystic change, and no hemorrhage within the tumor. The attenuation of the tumor was similar to muscle on postcontrast CT in all four cases. The extension of the tumor into the thyroid and loss of the fat plane between the tumor and the esophagus were demonstrated in all cases on CT. The signal intensity of the tumor was iso- to slight hyper-intense relative to muscle on T1-weighted images and of homogeneous high intensity on T2-weighted images. There was homogeneous dense enhancement of the tumor on T1-weighted postcontrast images	RR
	35	F	Neck mass, dyspnea		RR
	38	F	Neck mass		RR
	45	F	Dyspnea		RR
Idowu, 2004 (13)	68	F	Dyspnea	CT revealed a large, left-sided thyroid mass that extended to and completely encircled the trachea causing narrowing of the airway	RR
	60	M	Cough, hoarseness, and hemoptysis	CT scan showed involvement of the larynx and trachea with enlargement of the left thyroid lobe	Radiotherapy
Kukwa, 2014 (14)	17	F	Cough, hemoptysis, dyspnea and breathlessness	US revealed a 37×26 mm hypoechoic lesion between the left lobe and the trachea, with enlarged hypoechoic cervical nodes. CT confirmed a 34×37×50 mm tumor surrounding the trachea, infiltrating the two thyroid lobes and causing esophageal constriction	RR
Qi, 2016 (6)	46	M	Dysphagia and dyspnea	CT of the trachea showed tracheal compression and displacement with slight narrowing. Thyroid CT examination showed multiple thyroid nodules. The most prominent nodule was round and hypodense, and it was located in the right thyroid lobe. The nodule measured 84 HU, unenhanced, and 95 HU with slight enhancement on a contrast-enhanced scan	Local excision
Aldrees, 2016 (15)	47	F	Neck swelling, cough, shortness of breath, and hoarseness	US revealed a normal-sized right thyroid lobe, with a left lobe measuring 1.7×1.6×4.4 cm. CT revealed diffuse enlargement of the thyroid, more prominent on the left side, with significant tracheal compression and possible underlying infiltrative processes, but no cervical lymphadenopathy	RR
Al Khatib, 2017 (17)	54	F	Fullness in the neck, dyspnea, and dysphagia	US showed a left-sided solid soft tissue mass invading/ arising from the left lobe of the thyroid CT of the neck and chest showed a large heterogeneous soft tissue mass in the lower neck, inseparable from the left lobe of the thyroid, with retrosternal extension and shifting the trachea to the right side	Radiotherapy
Jikuzono, 2022 (16)	77	F	Dyspnea, hoarseness	CT and US revealed a tumor extending to the right thyroid lobe and peritracheal tissue, as well as severe stenosis of the lumen between the first and third tracheal rings	RR

F, female; NA, not available; RR, radical resection; M, male; CT, computed tomography; HU, Hounsfield units; US, ultrasound.

invasion (13). In the literature reviewed, nine patients underwent radical surgery, one patient underwent local excision, and two patients underwent radiation therapy. Our patient also underwent radical surgery with an uneventful postoperative recovery.

In our case, conventional ultrasound showed the lesion had unclear boundaries, irregular shape, and internal microcalcifications. CEUS showed heterogeneous hypo-enhancement in the central area of the lesion, and the texture of the lesion was hard on elastography. The lesion was adjacent to the peripheral dorsal membrane and closely related to the trachea. Based on the characteristics of multimodal ultrasound images, the lesion was considered to be a malignant tumor. Thyroid nodules are common in the population, but only about 5% of thyroid nodules are malignant (18). Papillary and follicular carcinomas are the most common thyroid cancers, accounting for more than 90% of all thyroid cancers (19). Ultrasound is currently the preferred imaging examination for thyroid diseases. Therefore, when the multimodal ultrasound shows that a thyroid nodule is malignant, we need to distinguish it from the common malignant tumors of the thyroid. Papillary thyroid carcinoma usually presents as hypoechoic nodules with microcalcifications, taller than wide, often associated with cervical lymph node metastasis, showing hypo-enhancement on CEUS and high stiffness on elastography (20,21). Follicular carcinoma usually presents as hypoechoic or isoechoic nodules with halo, and internal microcalcifications are rare. Some studies have indicated that follicular carcinoma shows hyper-enhancement on CEUS and soft texture on elastography (22-24). In conclusion, the multimodal ultrasound findings of TACC with thyroid invasion are not specific. When the multimodal ultrasound findings of thyroid nodules are malignant, different from the common thyroid tumors, and the nodules are closely related to the trachea, and the patient has clinical symptoms such as dyspnea, hoarseness, cough, and dyspnea, we should consider the possibility that the nodule is TACC with thyroid invasion.

Conclusions

TACC invading the thyroid manifesting as thyroid tumor is rarely reported. More literature is needed to report the sonographic manifestations of this disease. Multimodal ultrasound has shown some potential in the diagnosis of this disease and may be helpful for FNA and CNB. We need to consider the possibility of TACC with thyroid invasion

based on the patient's clinical presentation, ultrasound performance, and laboratory tests. Preoperative pathological examination and comprehensive imaging examination are necessary to help patients with better clinical management.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://gs.amegroups.com/article/view/10.21037/gS-23-485/rc>

Peer Review File: Available at <https://gs.amegroups.com/article/view/10.21037/gS-23-485/prf>

Conflicts of Interest: Both authors have completed the ICMJE uniform disclosure form (available at <https://gs.amegroups.com/article/view/10.21037/gS-23-485/coif>). Both authors report that this research was supported by National Natural Science Foundation of China (No. 82071940). The authors have no other conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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