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Case Report

Adenoid cystic carcinoma of trachea mimicking a thyroid follicular tumor: A case report and literature review [☆]

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ABSTRACT

Cervical tracheal adenoid cystic carcinoma (TACC) invading the thyroid gland is very rare and easily misdiagnosed as thyroid tumor, this paper reports a TACC invading thyroid in a 33-year-old woman who had been diagnosed as a thyroid follicular tumor in right lobe of thyroid by sonography and ultrasound guided fine needle aspiration in other hospital. She accepted surgical treatment in our hospital and was diagnosed as TACC by pathology, locally involving the thyroid. This paper presents the patient's clinical data, imaging findings, pathological diagnosis, treatment process and reviews the literature of TACC mimicking a thyroid tumor.

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Introduction

Adenoid cystic carcinoma (ACC) is a relatively rare malignant epithelial tumor that usually originates in the salivary glands, but it can also occasionally occur in the mammary glands, cervix, Bartholin glands, sinuses, skin, and even the larynx and trachea¹. TACC arises from glands in the tracheal and bronchial walls. The histological structure and biological behavior of TACC is similar to that of ACC found in salivary

glands, TACC commonly involves the cricoid cartilage, larynx, and subglottic region. Most of the TACC invading the thyroid gland mimicked thyroid tumors, especially thyroid follicular tumors [1]. Some imaging and pathologic features are helpful for differentiating TACC from thyroid tumors. Surgery remains the first choice of treatment for locally advanced TACC. Given the infiltrative properties of TACC and perineurial infiltrative activity, postoperative radiotherapy is often used to improve local control. In this report, we present a case of TACC mimicked thyroid follicular tumor.

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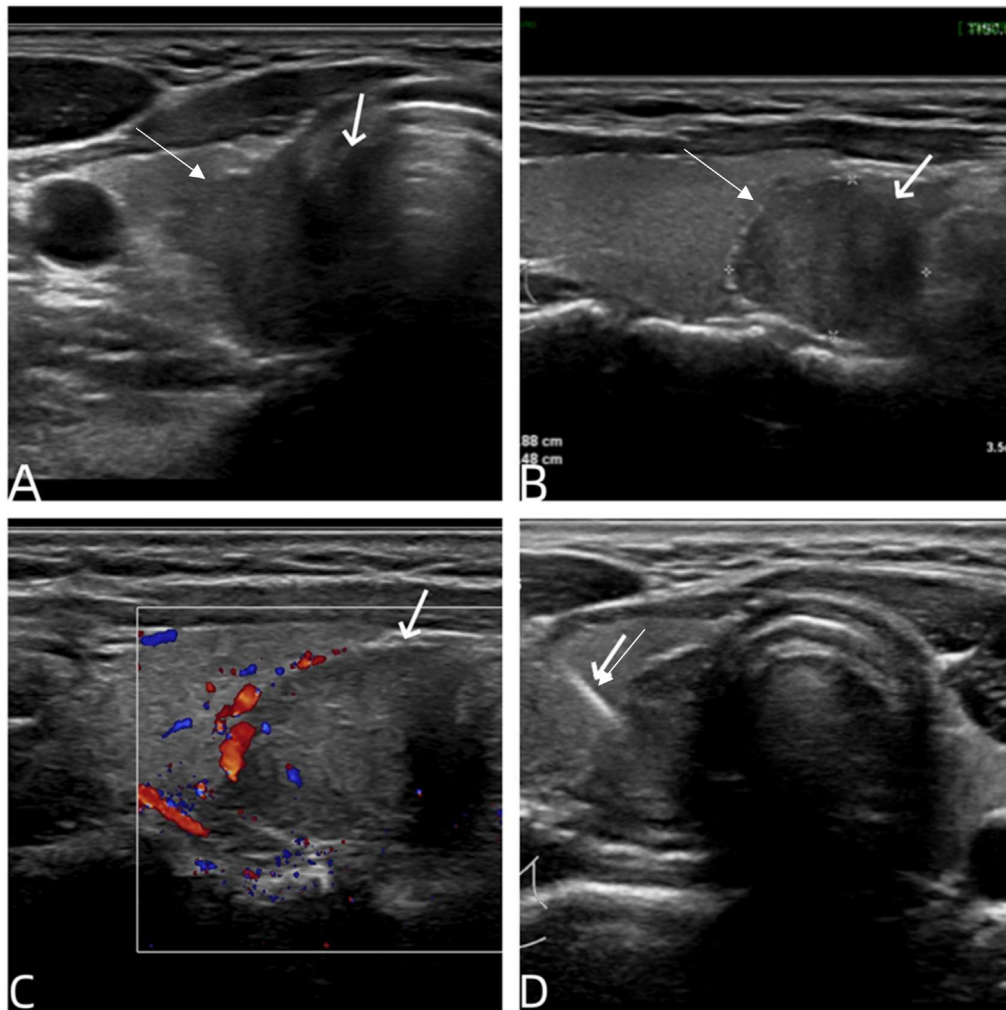


Fig. 1 – Ultrasonography. (A) there was a hypoechoic nodule (white arrow) in the right lobe of the thyroid, inseparable from trachea in the transverse section. **(B)** The nodule measured 1.9 × 1.5 cm and located in the lower part of the right lobe of thyroid in longitude section. There was a fine hyperechoic line between the nodule and the thyroid (white arrow). **(C)** There was peripheral strip blood flow and internal punctate blood flow in color Doppler flow imaging. **(D)** Ultrasound-guided fine needle aspiration biopsy. White arrow showed the needle.

Case presentation

A 33-year-old woman was admitted to our hospital with a nodule in the right lobe of thyroid detected by sonography accidentally, which was suspected to be a follicular tumor of the thyroid both by sonography and fine needle aspiration biopsy in another hospital. She denied symptoms of swallowing or breathing discomfort, hemoptysis, asthma, and so on. A tender nodule was palpated in the right thyroid on physical examination. All laboratory tests for thyroid including triiodothyronine, thyroxine, free triiodothyronine, free thyroxine, thyroid-stimulating hormone, thyroglobulin, thyrotrophin receptor antibody, thyroid peroxidase antibody, thyroglobulin antibody are normal.

Sonography showed a hypoechoic nodule located between the right lobe of the thyroid and the trachea which was poorly demarcated from the trachea, there was a hyperechoic line

between nodule and the thyroid, the Doppler sonography showed peripheral and internal blood flow. The patient underwent ultrasound guided fine needle aspiration biopsy again in our hospital (Fig. 1). Computed tomography (CT) imaging showed a hypodense mass located on the right wall and the posterior wall of trachea and close to esophagus, and there was no fat space between the mass and the right lobe of thyroid (Fig. 2). MR imaging of the neck showed a long T1 long T2 nodule in the right lobe of thyroid, which enhanced on contrast-enhanced scans and invaded membranous wall of trachea (Fig. 3). Fine needle aspiration cytology (Fig. 4) revealed abundant basal-like tumor cells arranged in a sieve-like pattern and surrounded ball-like mucous stroma, which was diagnosed as adenoid cystic carcinoma cells.

Obtaining the diagnosed of cytology, the patient underwent extended resection of tracheal tumor, tracheal sleeve resection (stepped-mode), right thyroid lobe and right recurrent laryngeal nerve resection. During the operation, a 2.3cm in

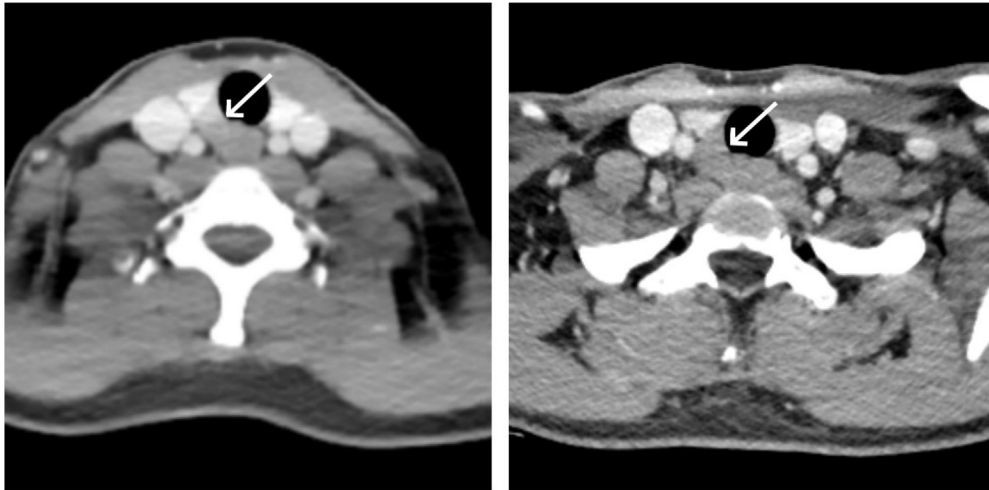


Fig. 2 – Enhanced CT images of trachea and thyroid. (A) A nodule with slight enhancement (similar to cervical muscle) (white arrow) was on the right side of the trachea, dorsal to the right lobe of thyroid. (B) The nodule (white arrow) invaded posterior wall of trachea and esophagus.

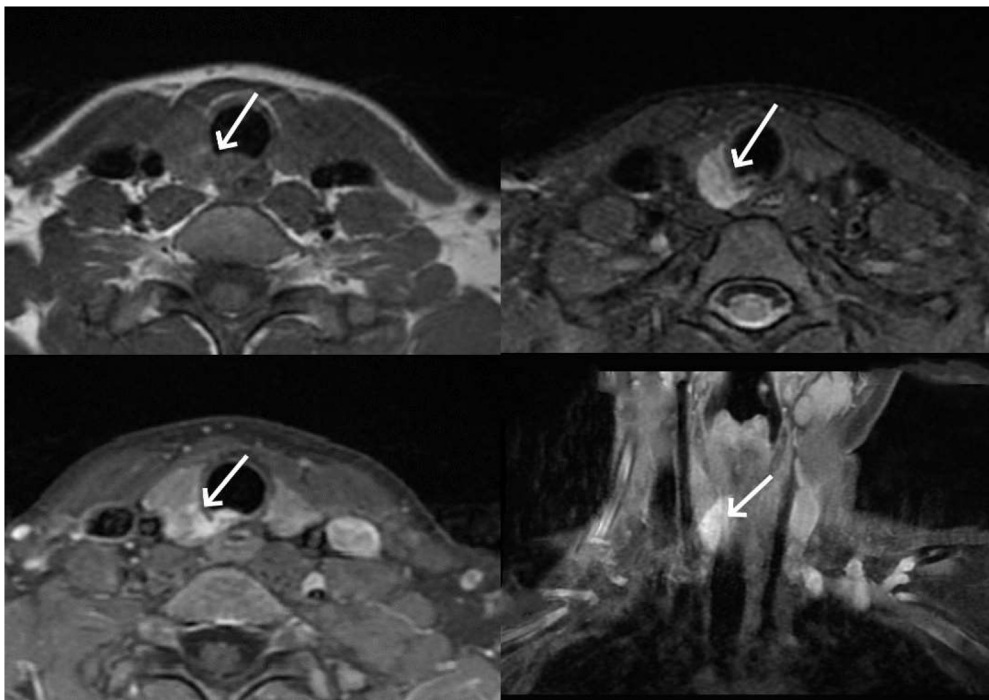


Fig. 3 – MRI images of the neck. (A) There was an irregular hypo-signal nodule (white arrow) in the right lobe of thyroid on T1WI transverse imaging. (B) The nodule (white arrow) was hyper-signal on T2WI/FS image. (C) The nodule (white arrow) was enhanced on contrast-enhanced T1WI transverse imaging and invaded the membranous part of the trachea.

length mass on the right wall of the trachea 1-5 rings was revealed, which invaded the membranous wall of trachea posteriorly and adhered to the esophageal muscularis and invaded the lower pole of the right lobe of the thyroid laterally and closely adhered to the recurrent laryngeal nerve and immediately adjacent to the cricoid cartilage. Post operation histopathology (Fig. 5) revealed a cribriform adenoid cystadenocarcinoma of the trachea, with a maximum diameter of 1.8

cm, invading the surrounding fibroadipose tissue and local thyroid tissue, nerve invasion was observed, and no clear vascular tumor thrombus was observed.

After operation the patient underwent 33 times of radiotherapy (radiation energy: 6Mv-X, dose: 69.96 Gy). Four months after the radiotherapy, the patient developed worsening dyspnea due to airway stenosis caused by radiotherapy, then she underwent a tracheotomy and a stage II trachea

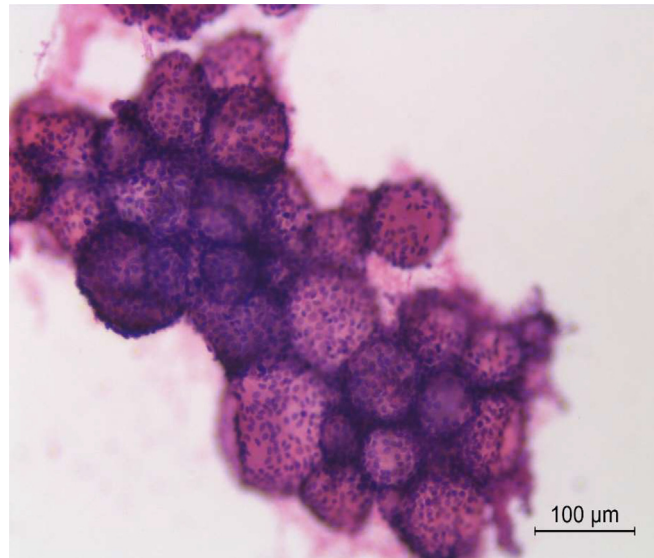


Fig. 4 – Cytological findings. Abundant basal-like tumor cells arranged in a sieve-like pattern and surrounded ball-like mucous stroma. Cytological diagnosis: adenoid cystic carcinoma cells (HE stain, Routine smear).

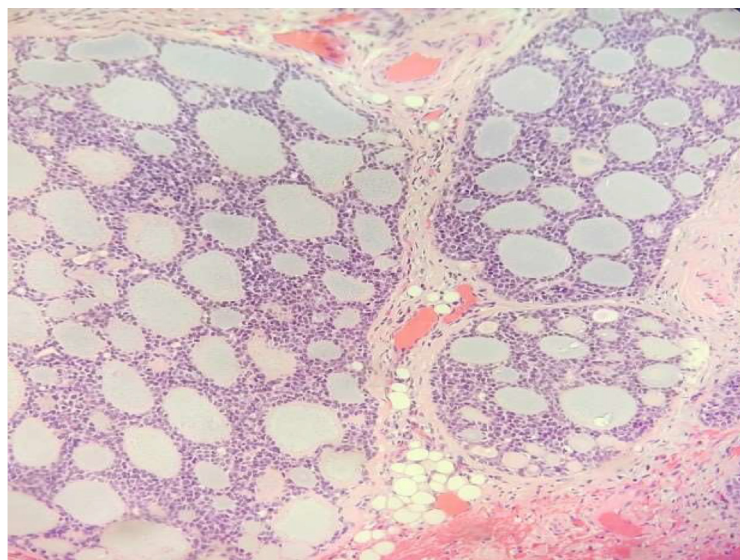


Fig. 5 – Histological findings. Adenoid cystic carcinoma of the trachea, cribriform type, involving the surrounding fibroadipose tissue and local thyroid tissue (200x HE).

reconstruction operation and there was no sign of tumor recurrence. The patient has been followed up for 17 months so far, and there is still no sign of local recurrence or distant metastasis.

Discussion

The majority of tracheal neoplasms are squamous cell carcinomas. Only 10% tracheal neoplasms are adenoid cystic carcinomas. Most TACCs are diagnosed in middle-aged individuals, TACC of the upper trachea most commonly involves the cricoid cartilage, larynx, and subglottic region, there were only

a few cases of TACC invade thyroid according to literature and when the tumor extended to the thyroid, it often presented clinically as a thyroid tumor [1]. Patients with TACC can present with slowly progressive respiratory symptoms such as polypnea, cough, hoarseness, hemoptysis, stridor, and neck pain [2].

As for our case, the nodule was found in thyroid by sonography and considered as follicular thyroid tumors by cytology in another hospital. Follicular thyroid carcinoma is the second most common thyroid malignancy tumor after papillary thyroid carcinoma [3]. Thyroid follicular tumor often presents as a hypoechoic/isoechoic round or oval nodule with a hypoechoic halo on sonography [4]. Our case of TACC presented as a single oval hypoechoic nodule located in right lobe of thyroid on ul-

trasound, which was easily misdiagnosed as thyroid follicular tumor. But there were some signs including a fine hyperechoic linear between the nodule and the thyroid, a convex margin to thyroid, the center of the nodule located outside the thyroid, the inseparable margin from trachea suggesting the nodule originated from the trachea rather than the thyroid.

CT examination is the commonest imaging method for tracheal tumor, which is good at display tracheal wall thickening and extraluminal extent of the tumor. TACC often shows a homogeneous mass encircling the posterolateral wall of trachea with thickened tracheal walls both on transverse and longitudinal CT. When the tumor invades the thyroid, it has a convex margin to the thyroid [5]. The CT examination for our case showed a mass encircling the posterolateral trachea, with a density close to muscle after enhancement, which is consistent with ACC.

MRI has high superior tissue characterization and shows the structure of the trachea more clearly. If MRI shows soft tissue signals in the tracheal cartilage or an intraluminal mass or a tumor surrounded trachea 180° or more, it strongly suggests a trachea tumor. In our case, MR showed the tumor protruding into the tracheal lumen at the membranous part of the trachea, suggesting the tracheal origin. It also showed tumor invasion of the thyroid clearly.

Cytology findings of ACC are usually a monomorphic population of basaloid cells in tight clusters with scant cytoplasm, acellular hyaline stroma in ball or cylinder-shaped also existed. Sometimes TACC can be misdiagnosed as follicular thyroid tumor because sample prepared improperly or the sample was mixed with thyroid tissue when the needle went through thyroid [6].

There are few literature reports describing TACC with thyroid invasion. Including our case, 14 cases of primary TACC with thyroid infiltration were reported [7–11]. According to Aldrees T there was another TACC misdiagnosed as thyroid follicular tumor [6]: the case presented as asthma and ultrasound revealed a normal-sized right thyroid lobe and slightly enlarged left thyroid lobe measured $1.7 \times 1.6 \times 4.4$ cm, both lobes exhibited heterogeneous echotexture, but left lobe was more prominent, which demonstrated increased vascularity with no discrete nodules. Computed tomography (CT) revealed diffuse enlargement of the thyroid, more prominent on the left lobe, with significant tracheal compression and possible underlying infiltrative processes. Fine-needle aspiration (FNA) of the thyroid reported findings consistent with follicular tumor. During operation frozen pathological revealed poorly differentiated carcinoma with adenoid cystic features in the tracheal wall and thyroid gland. And the final pathology confirmed the diagnosis of ACC. Therefore, ACC of the cervical trachea invades the thyroid gland and may present as a local lesion of the thyroid gland as we describe, or as diffuse changes in the thyroid gland as reported by Aldrees T.

as thyroid tumor. Careful analysis of the relationship between nodule and the thyroid and trachea is helpful for judging the origination of the tumor by using a variety of imaging methods including ultrasound, CT, and MRI. Ultrasound-guided FNA cytological examination findings of acellular hyaline stroma in ball or cylinder-shaped and basaloid cells are typical sign for TACC. Surgery is the first choice of treatment and requires completion of extended resection of the primary tumor, along with resection of the affected nerve and potentially invading tissue.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

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Conclusion

Upper tracheal TACC can directly invade the thyroid gland, form a nodule in the thyroid gland, and is easily misdiagnosed