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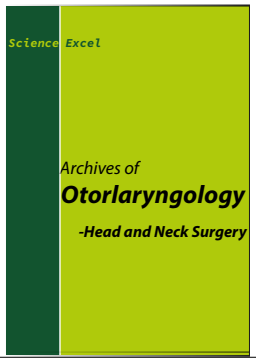
Case Report



Cystic Adenoid Carcinoma Of The Trachea: A Rare Case Of Asymptomatic Tracheal Tumor

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Abstract

Aim: To present the imaging appearance of tracheal adenoid cystic carcinoma according to the different imaging modalities (ultrasound, computed tomography and magnetic resonance imaging).

Method: We present the case of a 60-year-old patient, without symptoms guiding the diagnosis, in whom the diagnosis was suspected on CT scan imaging.

Results: The patient was explored by CT angiography of the supra-aortic trunks as part of the etiologic investigation of an ischemic vascular accident. The examination objectified a tissular mass centered on the tracheoesophageal axis lateralized to the left invading the laryngeal cartilages and the left lobe of the thyroid.

A cervical ultrasound was performed showing a heterogeneous hypoechoic area of the left lobe of the thyroid with no detected nodule. Fine needle aspiration of the suspicious area was performed, showing a tumoral lesion with basaloid cells evoking the diagnosis of adenoid cystic carcinoma.

In order to assess the tumor extension and the patient's operability, a cervical MRI was performed given its better resolution in contrast.

A bronchial fibroscopy was indicated showing a tracheal circumferential infiltrative stenosis with biopsies confirming the diagnosis.

Conclusion: Adenoid cystic carcinomas of the trachea are rare tumors. Tracheobronchial endoscopy combined with biopsies allow the diagnosis of certainty.

Cross-sectional imaging (CT and MRI) plays a key role in locoregional and distant extension assessment, thus guiding therapeutic management.

Introduction

Cystic adenoid carcinomas are low-grade tumors with submucosal locoregional extension.

They are often diagnosed late and present with nonspecific clinical features.

Case report

WWe report the case of a 60-year-old male patient with diabetes and hypertension, who presented with an ischemic stroke and, on Doppler ultrasound of the supra-aortic trunks, showed an atheromatous plaque in the left common carotid artery. Further investigation with a supra-aortic angioscan revealed a tissue process centered on the left tracheo-esophageal axis (Figures 1 and 2).

A cervical ultrasound was performed, revealing a heterogeneous hypoechoic area in the left lobe of the thyroid without any detectable nodules. A thyroid fine-needle aspiration biopsy was performed, which confirmed the presence of an invasive lesion with basaloid cells, suggestive of adenoid cystic carcinoma (Figure 3).

We performed a cervical MRI to assess tumor extension and operability of the patient. This examination revealed a tissue process centered on the posterior and left lateral walls of the first two tracheal rings, breaking the cartilaginous ring and adventitia at this level. It invades the laryngeal cartilages, the thyroid, and the cervical esophagus, extending into the prevascular retrosternal space (Figure 4).

A bronchoscopic examination was performed, revealing a circumferential infiltrative stenosis approximately 2-3 cm from the tracheal origin, causing a reduction of approximately 50% in its lumen.

Biopsies were taken at this site, confirming the presence of a carcinomatous proliferation consistent with adenoid cystic carcinoma.

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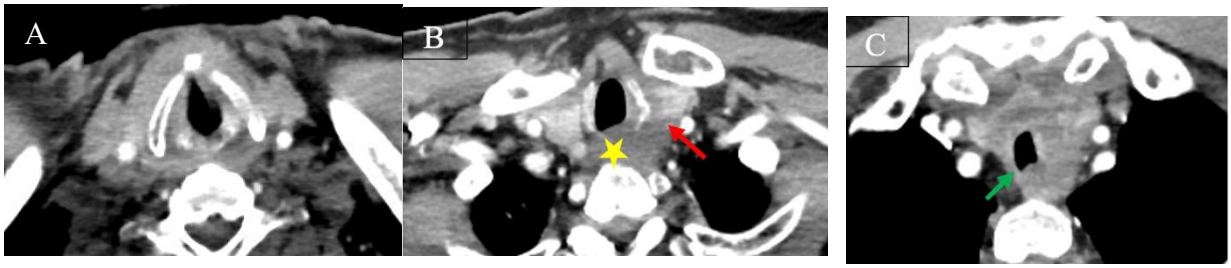

Figure 1.Axial CT scans centered on the cervical region with contrast-enhanced iodine injection.
A, B: Tissue process centered on the left tracheo-esophageal axis (yellow star) invading the cricoid cartilage, the inferior horn and left quadrilateral lamina of the thyroid cartilage, as well as the lower pole of the left lobe of the thyroid gland, extending to the thyroid isthmus (red arrow). C: It encases the trachea, which is laterally displaced to the right and reduced in caliber (green arrow).

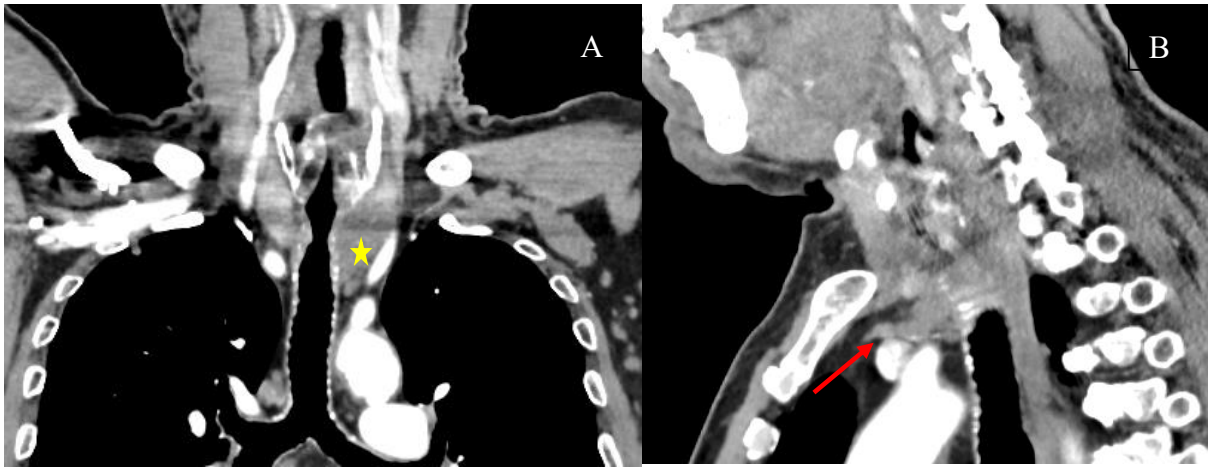

Figure 2.Coronal reconstruction in A and sagittal reconstruction in B of a cervico-thoracic CT scan with contrast-enhanced iodine injection.
The process extends downwards into the prevascular mediastinal space (red arrow), reaching the left innominate vein trunk and coming into contact with the left common carotid artery (yellow star) without obvious signs of invasion.

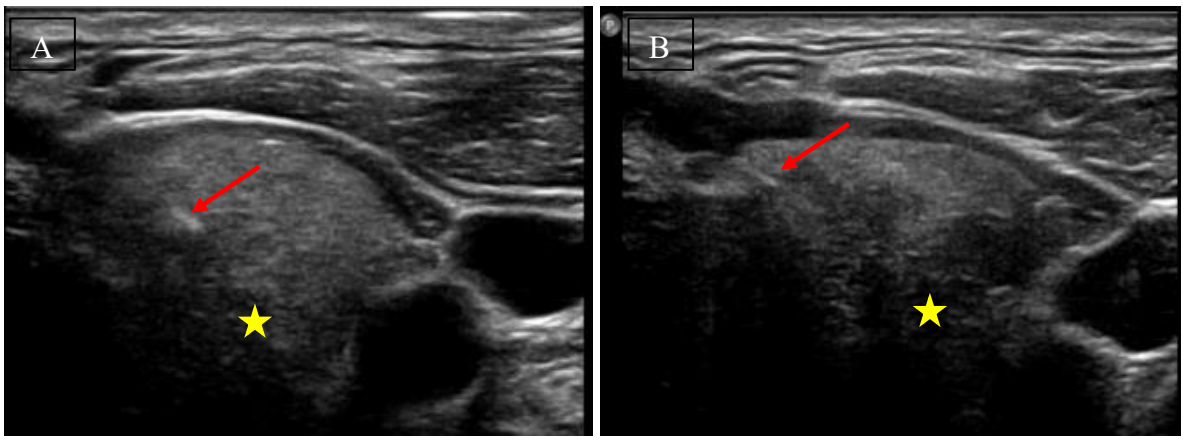

Figure 3.Transverse sections of a cervical ultrasound in A and B.
Heterogeneous hypoechoic area in the left lobe of the thyroid (yellow star). Fine-needle aspiration needle (red arrow).

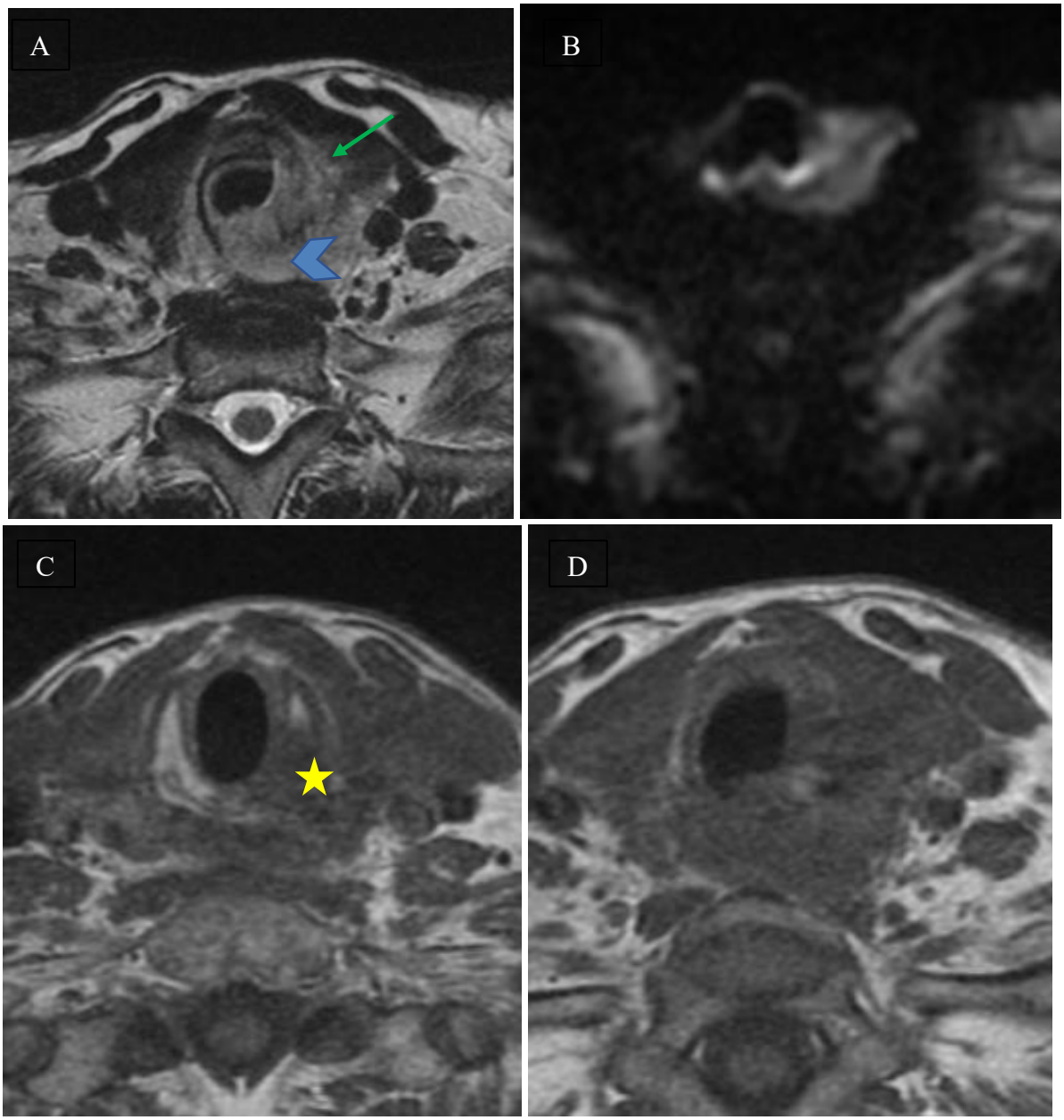

Figure 4.Axial slices of a cervical MRI with T2-weighted imaging in A, diffusion-weighted imaging in B, T1-weighted unenhanced imaging in C and D, and T1-weighted imaging with fat saturation and Gadolinium injection in E and F.

Tumor process showing heterogeneous hypointense signal on T1, intermediate signal on T2, hyperintense signal on diffusion, and intense heterogeneous enhancement after Gadolinium injection.

It invades the posterolateral aspect of the cricoid cartilage (yellow star), the lower horn and left lamina of the thyroid cartilage (red arrow), and the lower pole of the left thyroid lobe extending to the isthmus (green arrow).

It also invades the esophagus with tumor replacement of the mucosal signal (blue arrowhead).

Discussion

Primary tracheal tumors are very rare, accounting for less than 0.1% of all tumors. Around 90% of tracheal tumors are malignant, with squamous cell carcinoma being the most common type [1].

Adenoid cystic carcinoma (formerly known as cylindroma) is the second most common primary tracheal cancer, representing 5 to 10% of cases [2].

It was first described by Billroth in 1859 [3].

Adenoid cystic carcinoma is a malignant tumor that originates from the major or minor salivary glands in the head and neck region. It typically affects middle-aged individuals, with an equal sex ratio and no known risk factors (such as smoking or chronic alcoholism) [4].

The clinical presentation is nonspecific, characterized by a progressive cough and dyspnea. This often leads to initial differential diagnoses of asthma or chronic bronchopneumopathy [5].

The clinical symptoms are usually late in onset, with an

average time interval of more than 6 months from the first symptom. This is explained by the large diameter of the trachea and the slow growth of the tumor [6, 7].

Patients may also be asymptomatic, as in the case of our patient, and the diagnosis is often delayed due to the low-grade histological nature and slow progression of these tumors. Adenoid cystic carcinoma primarily affects the distal part of the trachea and main bronchi. Involvement of the lobar or segmental bronchi is exceptional [5, 8].

Typically, the tumor exhibits submucosal local extension with longitudinal and transverse progression along the posterior wall, related to the distribution of the accessory salivary glands located between the cartilage and the tracheal fibromuscular membrane. In advanced cases, invasion of adjacent structures can be observed [5, 8].

Lymph node metastases are uncommon (10 to 15%), while distant metastases occur in 15 to 40% of cases and preferentially affect the lungs, bones, brain, and rarely the liver.

Definitive diagnosis is made histologically through analysis of biopsies obtained during tracheobronchoscopic examination. However, cross-sectional imaging is necessary to establish a comprehensive assessment of local and regional extension due to the submucosal development of these lesions. Computed tomography (CT) can evaluate both the transverse and longitudinal local and regional tumor extension, as well as distant metastases (to the lungs, liver, brain, and bones).

On CT, adenoid cystic carcinoma appears as an endoluminal mass with tissue density, extending along the tracheal wall, circumferential thickening of the tracheal wall, or a tissue mass encircling the trachea with parietal thickening. Longitudinal extension is generally greater than transverse extension, with a predilection for involvement of the posterolateral tracheal wall [9]. In the extratracheal form, adenoid cystic carcinoma invades the thyroid gland and thyroid cartilage, as observed in our case [9].

Cervical MRI, with its superior contrast resolution, provides better assessment of local and regional extension, particularly submucosal extension. The curative treatment of adenoid cystic carcinoma is primarily surgical, performed in approximately half of the cases. Radiotherapy is indicated when complete or feasible resection is not possible, or in cases of local recurrence or lymph node involvement [10]. Non-resectable patients may benefit from palliative endoscopic treatment, providing symptom relief and improved quality of life.

Conclusion

Adenoid cystic carcinomas of the trachea are rare tumors. Tracheobronchoscopic examination coupled with biopsies allows for a definitive diagnosis. Cross-sectional imaging (CT and MRI) plays a crucial role in assessing the local and regional extent as well as distant spread, guiding therapeutic management.

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