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Tracheal Adenoid cyst carcinoma: Unusual cause of hemoptysis. A case report and review of the literature

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ABSTRACT

Adenoid cystic carcinoma (ACC) of the trachea is rare, it represents 1% of all respiratory tract cancers. ACC are the second most common primary malignant tracheal neoplasms after squamous cell carcinoma. Dyspnea, nonproductive cough, and hemoptysis are the common initial and atypical symptoms. We report an extremely rare case of ACC of proximal trachea, in a 28-year-old male who presented with a 6 month history of hemoptysis and dyspnea. Laryngoscopy and computed tomography revealed a posterior tracheal tumor 1.7 cm long axis without extra-tracheal extension and without significant lung injury. Biopsy confirmed the diagnosis of ACC. The patient was treated by surgical resection and end- to- end anastomosis followed by adjuvant radiotherapy. Six months follow-up of the patient did not reveal local recurrence or distant metastases. Clinical features, histological appearance and differential diagnosis as well as treatment are discussed.

Introduction

Adenoid cystic carcinomas (ACC) are the second most common primary malignant tracheal neoplasms after squamous cell carcinoma [1]. ACC of the trachea is a rare neoplasm originating from submucosal glands of the tracheobronchial tree [2]. Presenting symptoms can vary but generally include dyspnea, cough, stridor, wheezing and rarely with hemoptysis [3]. Surgical excision is the main stay of the treatment, with radiotherapy can be used as an adjuvant to it. In this paper we report an ACC of the trachea revealed by hemoptysis and the literature of tracheal ACC is reviewed.

Case report

We report a 28 year old Moroccan man, no smoking, no history that this particular. He suffering from progressive dyspnea for 6 months associated with recurrent hemoptysis low abundance.

The chest radiograph was normal. Bronchoscopy was performed and showed a polypoidal mass arising from the anterior wall of the trachea. A chest CT confirmed the existence of an anterior tracheal tumor 1.7 cm long axis without extra-tracheal extension and without significant lung injury. One biopsy was performed, it was very hemorrhagic and inconclusive and it was not renewed. A circular resection of the trachea carrying the 4 first rings allowed for resecting the entire tumor. Continuity tracheal was reconstructed with end-to-end anastomosis. The postoperative course was unremarkable.

Macroscopic examination of the piece shows an exophytic protruding tumor with a smooth surface, hemorrhagic appearance and soft consistency and pendant to the posterior wall of the trachea. Surgical margins were negative. **(Fig 1)**

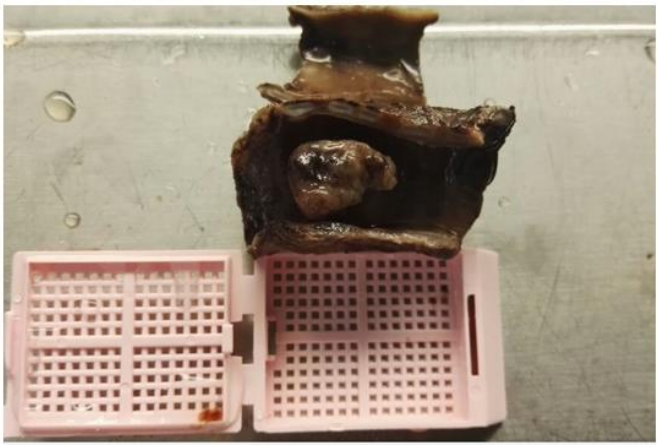

Figure 1. Photograph of the gross specimen obtained at tracheal resection shows a small polypoid mass arising from the anterior tracheal wall.

Microscopy showed infiltrating cribriform islands of tumor cells with small basophilic nuclei and lumina filled with eosinophilic material (Fig 2A). The pattern of infiltration extended from the mucosal surface through the cartilaginous tracheal ring and perineural invasion was prominent. (Fig 2B). The histomorphologic features and staining pattern were consistent with primary ACC of the trachea.

The patient also received external beam radiation therapy following surgery. The patient was on regular follow-up after the treatment. Follow-up of the patient after 6 months posttreatment did not reveal local recurrence or distant metastases.

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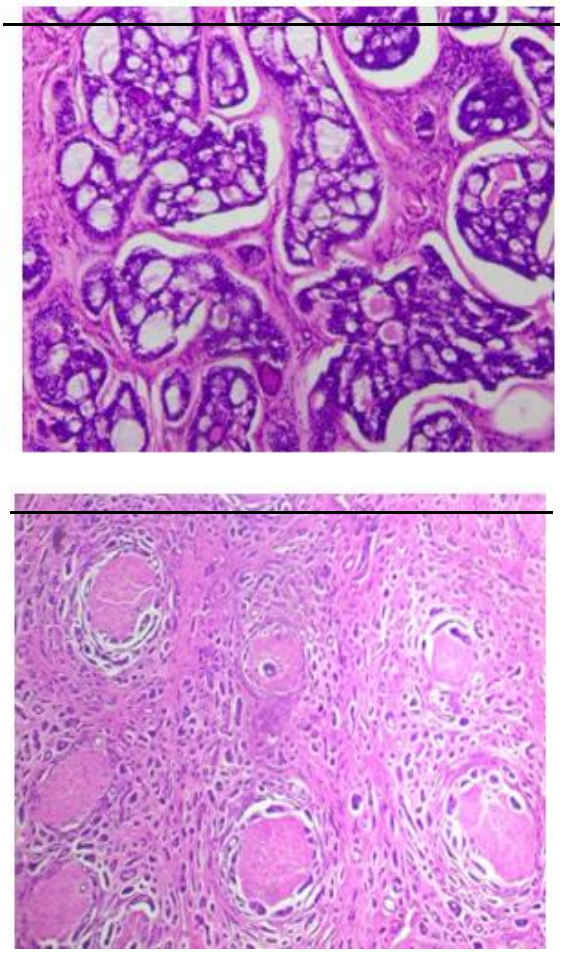

Figure 2.Adenoid cystic carcinoma.
(A): Proliferation composed of cylindromatous structures and trabecular structure. (Hematoxylin and eosin (HE) x 200)
(B): Numerous peripheral perineural invasion characteristic of the Adenoid cystic carcinoma. (Hematoxylin and eosin (HE) x 100)

Discussion

Primary malignant tracheal neoplasms are rare [4]. Squamous cell carcinoma is the most common type of malignant tracheal neoplasm, followed by ACC [5]. These two tumors represent approximately 86% of primary malignant tracheal tumors in adults. [6] ACC, formerly known as cylindroma, was first described by Billroth in 1859 [7].

Tracheal ACC demonstrates no racial or gender predilection, and no clear association with smoking has been proved [8, 9]. It is most common in patients in their 4th and 5th decades of life. [10, 11].

Symptoms associated with ACC are usually related to airway obstruction and these are dyspnea, cough, stridor, wheezing, change in voice and rarely hemoptysis. [12].

Radiologically, CT is considered to be the standard imaging modality for assessment of tracheal tumor; ACC has a tendency toward submucosal extension and on CT it appears as an intraluminal mass of soft tissue attenuation with extension through tracheal wall. The MRI does not have distinct advantage over CT in the evaluation of tracheal tumors. [13, 14].

Pathologically, at gross examination, ACC manifests as a poorly circumscribed, white-gray solid mass with infiltrating borders [15]. Microscopically, three patterns are seen; trabecular, cribriform and solid type. The cribriform pattern is most common consisting of uniform cells with relatively little cytoplasm arranged in well-defined nests of variable size. The cells in these nests are separated by well- defined cystic spaces containing a mucinous substance that stains strongly with alcian blue and weakly with Periodic Acid–Schiff (PAS) [16].

Pulmonary metastases are the most common and can remain asymptomatic for many years. Metastases to the brain, bone, liver, kidney, skin, abdomen, and heart have also been reported. [17].

Treatment of ACC includes surgery, radiotherapy, and combination of both. [18, 19]. Surgical resection followed by post-operative radiotherapy is the recommended treatment of choice. Radiotherapy is being used as an adjuvant after surgical resection or for unresectable tumors, medically inoperable cases or for palliation. [20].

Early diagnosis and treatment by surgery with radiation therapy provide significantly prolonged survival or even the possibility of complete remission.

Our patient is doing very well 6 months after surgery and postoperative radiotherapy without any evidence of local recurrence or distant metastases.

Conclusion

In summary, ACC of the trachea is a rare primary malignant tumor in adults. Nonproductive cough, and dyspnea are the common initial symptoms and rarely revealed by hemoptysis. ACC has an indolent course and good prognosis. Surgical resection followed by radiotherapy is widely recommended protocol for treatment and provides the best chance of prolonged survival.

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