



Case Report

Mucous Gland Adenoma of the Trachea: A Case Report Focusing on Endobronchial Treatment

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Central Message

Mucous gland adenoma of the trachea is a very rare benign tumor. We report the case of a 75 years old male, heavy smoker, complaining of cough, wheeze and shortness of breath. A large mass in the extra thoracic part of the trachea was revealed by fiberoptic bronchoscopy and resected using Argon Plasma Coagulation (APC) and electro surgery. Histopathology showed mucous gland adenoma. There are only 2 other cases of this type of adenoma, located in the trachea, in the current literature.

Keywords: Adenoma; Argon plasma coagulation; Bronchoscopy, Electro surgery, Mucous gland, Trachea

Introduction

Mucous gland adenoma is a rare benign tumor, originating from the mucous secretory glands of the large airway mucosa. The majority arise within the bronchi and lung parenchyma [1-3] but rarely in the trachea [4-6]. We report the third such case of mucous gland adenoma of the trachea.

Case description

A 75-year-old male, heavy smoker (180 pack/year), was referred to our hospital for evaluation of a mass in the extra thoracic segment of the trachea detected by chest Computed Tomography (CT). The patient experienced respiratory symptoms, such as cough, wheezing and shortness of breath for the last two months. Spirometry revealed a flow-volume loop pattern of fixed extra thoracic obstruction and Forced vital capacity (FVC) =1,88lt

(56% of the predictive values), Forced expiratory volume in 1 sec (FEV1) =1,71lt (68%), and FEV1/FVC =90%. Chest-X-Ray was normal, whereas the chest CT scan revealed the presence of an oval shaped mass, located in the upper tracheal lumen.

Patient underwent fiber optic bronchoscopy and endobronchial electro surgery. Premedication with 2mg midazolam i.v. was administered and lidocaine solution was sprayed in the upper respiratory tract prior to the insertion of the instrument. Supplemental oxygen was administered by the nasal cannula to maintain the oxygen saturation above 95.0%. The endoscopic procedure was performed with flexible fiberoptic bronchoscope Olympus BF 1T180 with a 2.8 mm working channel. The instrument was introduced nasally and the entire procedure was recorded and stored in electronic form. Endoscopic procedure revealed the presence of a well-defined large broad-based mass, with a smooth surface and marked vascularity, located 1,5centimeters from the vocal cords, obstructing the upper tracheal lumen. (Figure 1).



Figure 1: Endoscopic view of the endotracheal mass.

The rest of the lumen of the trachea was normal as well as, the mucosa till the carina. Argon Plasma Coagulation (APC) was performed for control of bleeding prior to definitive resection. Then, the mass was completely resected using a single-use polyloop Olympus device. A 1,8cm diameter mass was resected and send for histopathological examination. The entire procedure was well tolerated by the patient and 0,25mg anaxate was administered at the end of the procedure in order to reverse the midazolam effect. The duration of the entire procedure was 22 min and it was well tolerated by the patient. No significant complications observed. The patient showed immediately complete remission of symptoms, he left the hospital the same day and two years later he is free from respiratory symptoms.

Macroscopic examination of the resected specimen revealed an elastic and well-circumscribed whitish solid mass, 1.8 cm in maximum diameter. Microscopically, the tumor consisted of arranged cysts and adenoid glands lined by bland cylindrical, cuboidal and flattened cells without cytological atypia. Focally in the cylindrical cells oncocytes characters were observed. Many small cystic spaces were filled with mucoid material. Focally were present slightly dilated adenoid formations containing mucous. The intervening stroma consisted of delicate connective tissue. The tumor was well circumscribed. The external surface lined by respiratory epithelium, with absence of cellular atypia and

presence of foci of incomplete squamous metaplasia. In the sub epithelial layer an area of low-to-moderate grade of non-specific chronic inflammation was observed. The mucoid material of the cysts and the cytoplasm of several cells of the glands were positive for PAS stain. (Figure 2). Immunohistochemically the tumor cells were positive for CK7, a few cells were positive for TTF-1 and all the tumor cells were negative for P40. Positivity for TTF-1 and P40 was observed in the myoepithelial cells (Figure 3) Thus, the diagnosis of a mucous gland adenoma was made.

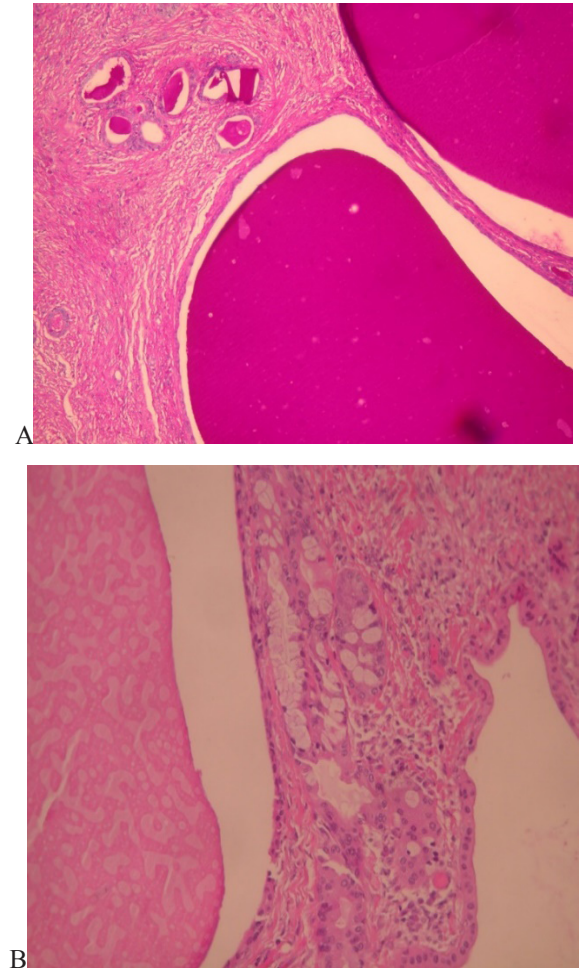


Figure 2: A: Cysts filled with PAS positive mucoid material. (PAS stain X100); B: Cyst filled with mucoid material and mucous secreting glands. (H+E x100).

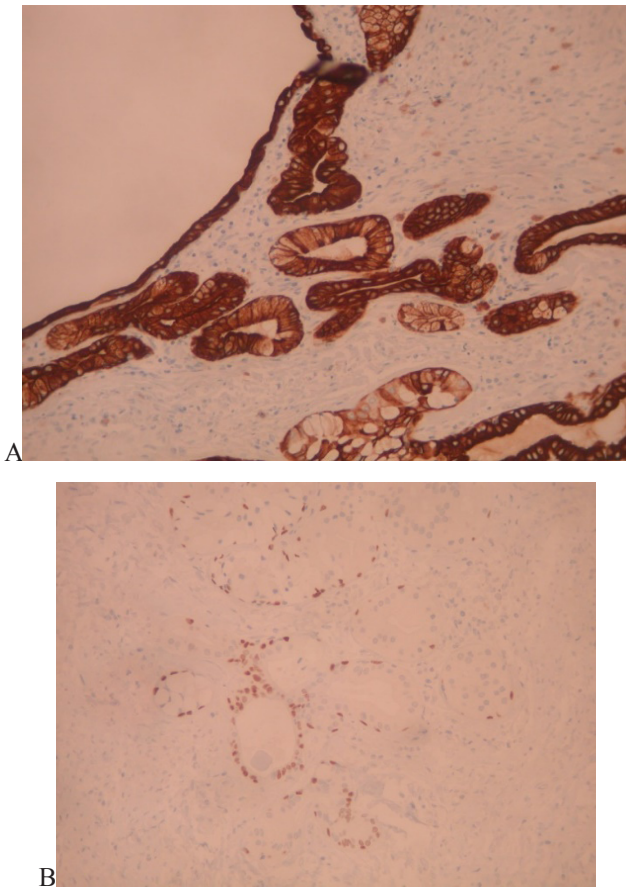


Figure 3: A: Epithelial cells positive for CK7, Immunostain x 200; B: Few epithelial but most myoepithelial cells positive for TTF-1, Immunostaining x 200.

Discussion

Mucous gland adenoma is a rare benign tumor originating from the mucous secreting glands of the larger airway mucosa. This tumor was first reported in 1882 by Muller as a pathologic entity separate from carcinoma of the lung and was first named bronchial adenoma arising from mucous gland [1]. The majority of the cases are seen in the bronchus, and more rarely in the trachea or peripheral airways. To the best of our knowledge, only 2 cases located in the trachea have been reported in the current literature and only six 6 cases located in the periphery of the lung [4-9].

Macroscopically, mucous gland adenoma appeared as a round, smooth, exophytic mass. Histologically, this type of tumor contains glands, acini, tubules, and even dilated mucus-containing cysts. Occasionally, a papillary pattern can be observed. The lining cells vary from cuboidal cells to tall columnar cells with mild cellular atypia. Stratified epithelia are not prominent and papillary proliferation is absent. The tumor often protrudes

into the bronchial lumen and is covered by the normal tracheal epithelia. Bronchoscopic treatment of endobronchial tumors can be accomplished in several ways [10,11]. These include electrocautery, cryotherapy, Argon Plasma Coagulation (APC), and laser (light amplification by stimulated emission of radiation) using Neodymium-Yttrium-Aluminum-Garnet (Nd:YAG), Neodymium-Yttrium-Aluminum-Perovskite (Nd:YAP), carbon dioxide lasers and others. Electrocautery involves tissue destruction using an electric current. It is best suited for small intraluminal tumors in early stages with curative intent. It has risk of intraluminal bleeding and perforation. APC, on the other hand, is a mode of noncontact electro-coagulation that involves forming plasma at the tip of a probe which produces coagulative necrosis in the targeted tissue. APC is useful in targeting lesions at sharp angles and provides excellent hemostasis. Laser therapy is useful for tumor debulking and among other used laser types, Nd:YAG is the most commonly used. Nd:YAG Laser has an invisible beam with wavelength that lies in the infrared region with depth up to 10 mm (compared to 1-5 mm for APC). With the exception of the carbon dioxide laser that requires rigid bronchoscopy, most other laser types can be used through the flexible bronchoscope [11,12].

In this case, the patient experienced symptoms such as cough, wheezing and shortness of breath for two months. The flow-volume presented inspiratory and expiratory limb of the loop both flattened showing the presence of fixed obstruction of the upper airways. The chest x-ray was not diagnostic but the tumor was detected by the chest CT scan [9]. We highlight the successful management of mucous gland adenoma via the less invasive outpatient-based approach. In fact, using flexible fiberoptic bronchoscopic endobronchial electro surgery the tumor was completely resected from the trachea releasing the airway with immediate remission of symptoms. It was a procedure well-tolerated by the patient who had no complication, didn't require hospitalization after the procedure and was sent home the same day. At two years follow up the patient is free of respiratory symptoms.

Moreover, benign glandular tumors of the tracheobronchial tree are very rare and unfortunately remain out of the limelight of pulmonary medicine. They produce symptomatic bronchial obstruction, and the clinical presentation may be mistaken for asthmatic disorder [8] or chronic obstructive pulmonary disease for long periods of time. Other nonspecific observations such as a chronic cough, unilateral wheezing, hemoptysis, or pulmonary infections can also be present for a long time before a correct diagnosis is made. Management of these tumors is the complete excision and requires a multi-disciplinary approach. The interventional pulmonologist, radiologist, anesthesiologist and pathologist play a crucial role in the management of these tumors. We highlight the necessity of an in-depth workup in patients presenting with signs of upper airway obstruction and such an unusual differential diagnoses should be considered.

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