

# Right main bronchial pleomorphic adenoma

## A case report and literature review

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### Abstract

**Rationale:** Tracheobronchial benign tumors are uncommon; particularly, bronchial pleomorphic adenoma is one of the rarest benign tumors that develop in bronchus (only 7 reported cases, among which only 4 cases of pleomorphic adenoma were seen arising from right main bronchus).

**Patient concerns:** In this report, a 38-year-old woman suffered from progressive shortness of breath for 5 years due to right main bronchial pleomorphic adenoma.

**Diagnoses:** The patient was diagnosed as right main bronchial pleomorphic adenoma based on chest computed tomography enhanced scan, bronchoscopy, and histological examination.

**Interventions:** An electrosurgical snare was performed to resect the neoplasm and several APC were administered at the sites of the resection to provide hemostasis and further coagulate for the residual neoplasm.

**Outcomes:** The patient was free of symptoms and the lumen of right main bronchus was clear during the follow-up period for 10 months without any procedure-related complications.

**Lessons:** Bronchial pleomorphic adenoma is extremely rare, however, we should take it into consideration if a patient suffered from shortness of breath without an exact cause.

**Abbreviations:** CT = computed tomography; APC = argon plasma coagulation; NBI = narrow band imaging.

**Keywords:** airway obstruction, bronchial pleomorphic adenoma, interventional therapy, location and therapy

### 1. Introduction

Pleomorphic adenoma is the most common type of tumor in salivary glands; however, tracheobronchial pleomorphic adenoma is extremely uncommon.<sup>[1]</sup> To the best of our knowledge, only 44 cases have been reported,<sup>[2–5]</sup> among which only 7 cases of pleomorphic adenoma were seen arising from main bronchus (4 cases arising from right main bronchus and 3 cases arising from left main bronchus). A patient who suffered from progressive shortness of breath for 5 years due to right main bronchial pleomorphic adenoma was reported in this article. An electrosurgical snare and argon plasma coagulation (APC) with a flexible bronchoscope, which was reported in using to treat

bronchial pleomorphic adenoma only once before, was used to resect the bronchial neoplasm.

### 2. Case description

The study protocol was reviewed and approved by the Institutional Review Board and the Ethics Committee of the Second Affiliated Hospital of Fujian Medical University, Fuzhou, China. Informed consent was obtained from the patient for publication of this case report and accompanying images.

A 38-year-old woman was referred to our department due to progressive shortness of breath for 5 years. On physical examination, sound of right lung was decreased. Laboratory tests including blood routine examination, biochemical examination, and tumor markers are all unremarkable. A chest computed tomography (CT) enhanced scan showed a neoplasm (oval, major axis 1.42 cm and minor axis: 0.96 cm) confined to the inner wall of right main bronchus (Fig. 1A). Subsequent flexible bronchoscopy revealed a pink, broad-based, polypoid neoplasm with branching blood vessels on the smooth surface arising from the left lateral wall of right main bronchus and nearly completely obstructing its lumen (Fig. 1B). A therapeutic flexible bronchoscopy was performed in an operating room and the bronchial neoplasm was resected through combining an electrosurgical snare and APC after signing the informed consent. Nearly total patency of the bronchial lumen was established after the bronchoscopy and the obstruction of the right main bronchus was resolved (Fig. 2). The shortness of breath was markedly improved without any procedure-related complications. The histological examination of the resected specimens showed trabecular and islands of epithelial and myoepithelial cells in a myxoid matrix and chondroid matrix (Fig. 3). On the basis of the findings of the histological examination, a definitive diagnosis of pleomorphic adenoma was made.

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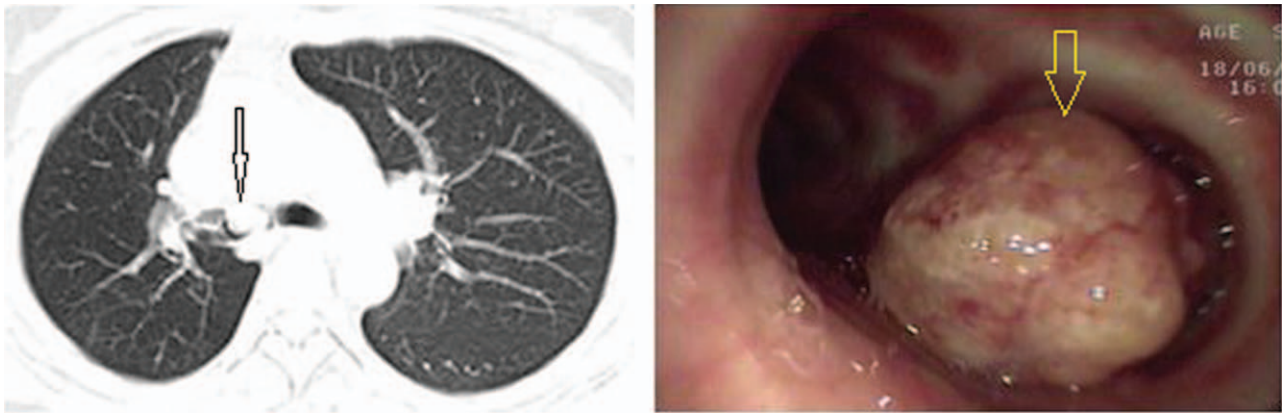
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**Figure 1.** Preoperative situation. A, Chest computed tomography (CT)-enhanced scan revealed an endobronchial mass located at the beginning of right main bronchus. B, Bronchoscopy revealed a polypoid neoplasm with branching blood vessels on the surface involving right main bronchus.

Repeated chest CT enhanced scan and bronchoscopy using narrow band imaging (NBI) guidance were performed on the 33rd postoperative day. The lumen of right main bronchus was clear on the chest CT enhanced scan and no regrowth of pleomorphic adenoma was found since no vascular abnormalities suggestive of neoplastic process were seen on the NBI (Fig. 4A,B). No obvious abnormalities in the lumen of right main bronchus were found on the latest chest CT enhanced scan and bronchoscopy performed 3 months later (Fig. 5A,B). The patient was free of symptoms during the follow-up period for 10 months.

### 3. Review of the literature

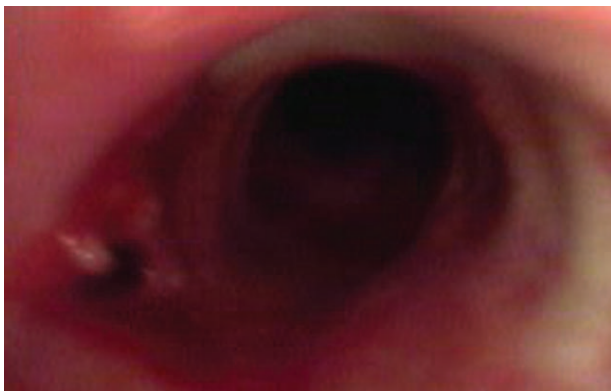
Literatures published in the latest 20 years were reviewed and the location and therapy of tracheobronchial pleomorphic adenoma were summarized (Table 1). In Table 1, the cases are separated into 2 groups based on the therapy methods.

### 4. Discussion

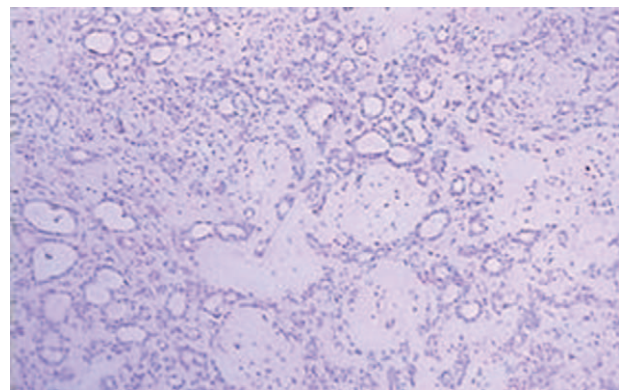
Benign tumors originating from the tracheobronchial tree are very uncommon, particularly, bronchial pleomorphic adenoma is one of the rarest benign tumor develops in the bronchus that consists of neoplastic myoepithelial cells mixed with neoplastic ducts and stroma.<sup>[6,20]</sup> The incidence, etiology, and prognosis are

still unclear due to its rarity.<sup>[21]</sup> No evident sex predominance, the age is usually varied from 26 to 76 years as reported in literatures though 2 additional cases have been reported, one is an 8-year-old boy<sup>[13]</sup> and the other is a 15-year-old boy.<sup>[11]</sup> Pleomorphic adenoma always occurs in the main or secondary bronchus and patients may have symptoms and signs such as cough, dyspnea, wheezing, repeated pulmonary infections, stridor, and shortness of breath owing to airway obstruction.<sup>[7,16]</sup> The symptoms and signs are various depending on the site and size of the tumor.<sup>[7]</sup> Because of the slow-growing nature of the tumor and the lack of specific symptoms and signs in this condition, the symptoms and signs may be misdiagnosed as bronchial asthma or chronic bronchitis and the proper diagnosis is usually delayed as well.<sup>[14]</sup> In addition, the tumor may not be discovered on thorax radiographs, accordingly, chest CT and bronchoscopy play a crucial role in diagnosis.<sup>[3]</sup> Same as described in literatures, our patient whose tumor was detected incidentally by chest CT had a 5-year history of progressive shortness of breath before the definite diagnosis of bronchial pleomorphic adenoma was made.

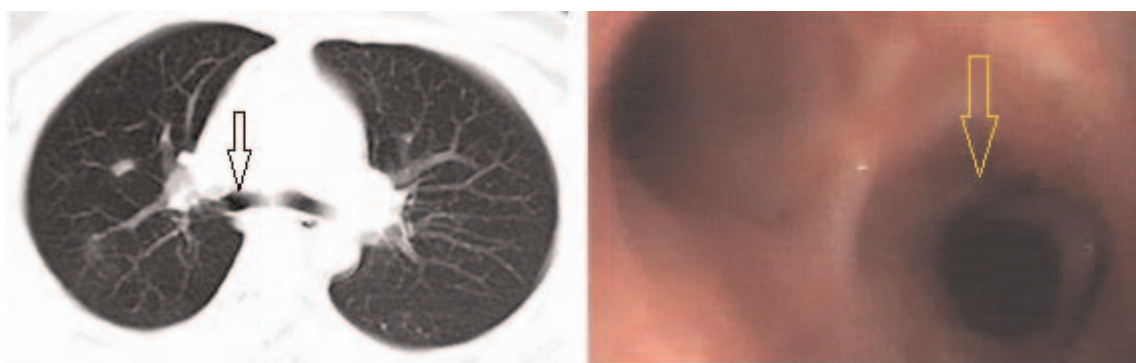
The histological characteristics of bronchial pleomorphic adenoma are similar to salivary gland tumors except that the ducts are relatively sparse.<sup>[16]</sup> Microscopic findings show sheets, trabecular or islands of epithelial and myoepithelial cells in a myxoid or chondroid matrix.<sup>[22]</sup> The most difficult differential diagnosis of pleomorphic adenoma is adenoid cystic carcinoma.



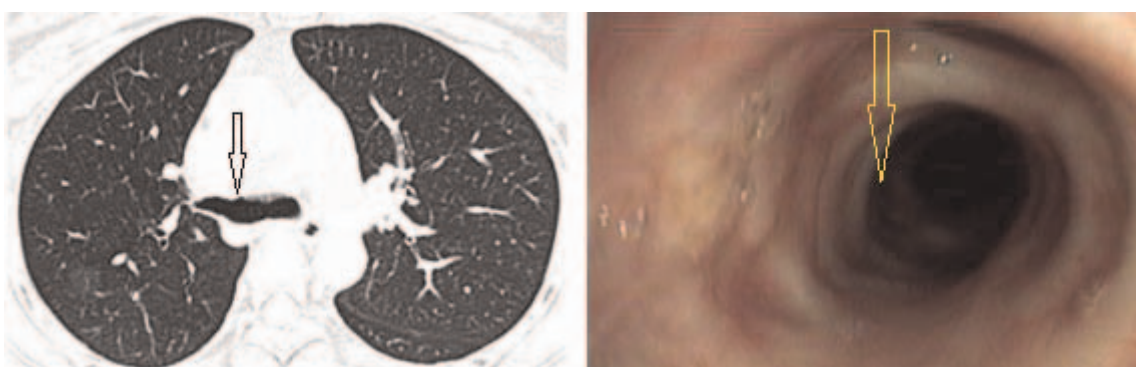
**Figure 2.** Nearly total patency of the bronchial lumen was established and the obstruction of the right main bronchus was resolved after therapy.



**Figure 3.** Histological finding of the neoplasm. An encapsulated biphasic lesion consisting of a mixture of epithelium and stroma with areas of hyalinization (40 $\times$ ).



**Figure 4.** Conditions 33 days after operation. A, The lumen of right main bronchus was clear. B, No vascular abnormalities suggestive of neoplastic process were seen on the narrow band imaging (NBI).



**Figure 5.** Conditions 3 months after operation. A, No obvious abnormalities in the lumen of right main bronchus. B, No regrowth of pleomorphic adenoma was found.

**Table 1**

**The location and therapy features of patients with tracheobronchial pleomorphic adenoma reports previously.**

Location	Therapy
Distal trachea	Endoscopic resection
Upper one-third of the trachea	Tumor was resected with rigid forceps and APC <sup>[5]</sup>
Left main bronchus	Tumor was resected with APC <sup>[4]</sup>
Distal trachea	Tumor was resected with electrocautery and rigid bronchoscopic coring <sup>[7]</sup>
Left main bronchus	Tumor was resected with electrocautery and rigid bronchoscopic coring <sup>[7]</sup>
Right main bronchus	Tumor was resected with electrocautery and rigid bronchoscopic coring <sup>[7]</sup>
Mid one-third of the trachea	Tumor was removed with diathermy snare <sup>[9]</sup>
	Tumor was excised with cold instruments <sup>[11]</sup>
Trachea	Surgical resection
Mid one-third of the trachea	Right thoracotomy with segmental resection and end-to-end anastomosis <sup>[3]</sup>
Left main bronchus	Collar incision with partial sternotomy and end-to-end anastomosis <sup>[2]</sup>
Lower one-third of the trachea	Wedge bronchiectomy <sup>[10]</sup>
Lower one-third of the trachea	Tracheal resection and end-to-end anastomosis <sup>[11]</sup>
Distal trachea	Right thoracotomy with circumferential resection and end-to-end anastomosis <sup>[12]</sup>
Trachea	Tracheal reconstruction <sup>[13]</sup>
Upper one-third of the trachea	Segmental tracheal resection and end-to-end anastomosis <sup>[14]</sup>
Distal trachea	Segmental tracheal resection and end-to-end anastomosis <sup>[15]</sup>
Distal trachea	Tracheal wedge resection <sup>[16]</sup>
Mid one-third of the trachea	Primary anastomosis performed through right thoracotomy <sup>[17]</sup>
Hilum of the right lung	Right thoracotomy incision and end-to-end anastomosis <sup>[18]</sup>
	Lobectomy <sup>[19]</sup>

APC=argon plasma coagulation.

Tumor cell nests of adenoid cystic carcinoma are sharply separated from the stroma.<sup>[2,3]</sup> Immunohistochemical staining may facilitate the correct diagnosis,<sup>[22]</sup> which may reveal the myoepithelial cell components are positive for cytokeratin, p63, and S-100 protein as well as ductal epithelial cell components are positive for cytokeratin, epithelial membrane antigen, secretory component carcinoembryonic antigen, lysozyme, and gross cystic disease fluid protein-15.<sup>[14,15]</sup> The most frequently expressed cytokeratin-19 is positive in the luminal cells of the tubular structures.<sup>[2,3]</sup> Such a routine study of tumor genetic expressions in all new patients will contribute to characterize the biologic profile and may play a role in predicting the frequent recurrence of the tumor.<sup>[22]</sup>

Owing to the rare occurrence, standard management for bronchial pleomorphic adenoma has not been established. In general, radical resection should be considered first due to the potential recurrence. Surgical tumor resection and airway anastomosis was suggested to be the optimal therapeutic choice in many literatures,<sup>[13,14]</sup> meanwhile, with the rapid development of interventional pulmonology, endoscopic resection has become a kind of curative and useful therapy for bronchial pleomorphic adenoma.<sup>[13]</sup> The combination of an electro-surgical snare and APC is one of the safest and most effective interventional techniques and whose immediate effect is now widely accepted.<sup>[13]</sup> In our case, we successfully performed an endobronchial resection of the tumor with a combination of an endobronchial electro-surgical snare and APC using a flexible bronchoscope. Although the last follow-up bronchoscopy suggested no tumors reoccurred, detailed genetic investigation and long-term follow-up are still indispensable due to the potential reoccurrence and malignant transformation.<sup>[12,22]</sup>

In conclusion, bronchial pleomorphic adenoma is a rare disease, which may lead to symptoms and signs such as cough, dyspnea, wheezing, and shortness of breath owing to bronchus obstruction. Chest CT and bronchoscopy can clearly reveal the tumor; microscopy and immunohistochemical staining are beneficial to the definite diagnosis. Although surgical radical resection is the optimal therapy due to the low possibility of recurrence, endoscopic resection is also a workable alternative method. Because of the low possibility of recurrence and potential malignant transformation, careful follow-up after resection procedure is necessary. Further studies with additional cases are needed to define the clinical course, biologic behavior, and prognosis of bronchial pleomorphic adenoma.

### Author contributions

**Conceptualization:** Zhixing Zhu, Dongyong Yang.

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**Writing – original draft:** Zhixing Zhu, Xihua Lian, Dongyong Yang.

**Writing – review and editing:** Zhixing Zhu, Xihua Lian.

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