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Contents

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World Journal of Clinical Cases

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Contents

World Journal of Clinical Cases

Semimonthly Volume 8 Number 23 December 6, 2020

Contents

World Journal of Clinical Cases

Semimonthly Volume 8 Number 23 December 6, 2020

Contents

ABOUT COVER

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CASE REPORT

Pleomorphic adenoma of the trachea: A case report and review of the literature

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Author contributions: Liao QN and Yu HP were the patient's respiratory physicians; Chen SB performed the radiological diagnosis and contributed to manuscript drafting; Fan HZ performed the pathological diagnosis and contributed to manuscript drafting; Liao QN, Fang ZK, Chen LC, Wu XP, and He X reviewed the literature and contributed to manuscript drafting; Liao QN, Fang ZK, Chen SB, and Yu HP were responsible for the revision of the manuscript for important intellectual content; all authors issued final approval for the version to be submitted.

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Abstract

BACKGROUND

Pleomorphic adenoma (PA) is the most common benign tumor that occurs in the salivary glands; however, tracheobronchial PA is rarely observed. To the best of our knowledge, fewer than 50 cases have been reported in the literature. We report a 49-year-old woman who had been treated for asthma for 2 years before being diagnosed with PA of the trachea.

CASE SUMMARY

A 49-year-old woman was referred to our hospital due to dyspnea upon exertion and chronic cough with wheezing for 2 years. Laboratory tests showed an elevated white blood cell count, absolute neutrophil count, and percentage of neutrophils. A chest computerized tomography scan showed a well-defined, soft-tissue density lesion measuring 2.4 cm × 2.1 cm in the lower trachea. Flexible bronchoscopy revealed that nearly 90% of the tracheal lumen was obstructed. The histopathological and immunohistochemistry features suggested PA of the trachea. Furthermore, we review the characteristics of 29 patients with tracheobronchial PA over the last 30 years.

CONCLUSION

Tracheobronchial PA occurs without gender predominance, mostly in the lower or upper trachea, and has a low recurrence rate. The median age at diagnosis is 48 years. The most common symptoms are cough, stridor, dyspnea, and wheezing.

Key Words: Pleomorphic adenoma; Trachea; Bronchoscopy; Review; Diagnosis; Case report

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Core Tip: Pleomorphic adenoma of the trachea is a rare benign tumor with slow growth. However, no standards for management have been established, and the clinical course has not yet been defined. In this study, 29 cases of tracheobronchial pleomorphic adenoma are reviewed with regard to the most common symptoms, clinical course, and treatment. For early and accurate diagnosis, chest computerized tomography and bronchoscopy should be performed initially in suspected cases.

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INTRODUCTION

Pleomorphic adenoma (PA) is an unusual type of salivary-gland neoplasm that occurs in the trachea[1]. The tumor is composed of recognizable epithelial tissue mixed with mucoid, myxoid, and chondroid tissues, which can also be observed in the soft palate, hard palate, upper lip, nasal septum, nasopharynx, orbital area, lower eyelid, buccal mucosa, cheek, and external auditory canal[2]. To the best of our knowledge, fewer than 50 cases have been reported[3-6]. Due to the lack of early specific symptoms, PA of the trachea is usually misdiagnosed as asthma[6-9]. In addition, cases of PA can progress to malignant tumors[10]. We present a case of PA of the trachea that was successfully treated by bronchoscopic interventions.

CASE PRESENTATION

Chief complaints

Dyspnea upon exertion and chronic cough with wheezing for 2 years.

History of present illness

A 49-year-old woman was referred to our hospital for dyspnea upon exertion and chronic cough with wheezing for 2 years. The above symptoms worsened with white mucus sputum for the past one week with no complaints of fever, chest tightness, chest pain, or hemoptysis.

History of past illness

The patient was previously diagnosed with asthma and treated with inhaled glucocorticoids for 2 mo.

Personal and family history

There was no history of tobacco use, and the patient denied having a personal or family history of other diseases.

Physical examination

In the physical examination, lip cyanosis, three depression signs (suprasternal fossa, supraclavicular fossa, and intercostal space), and expiratory and inspiratory wheezing were observed, and the sound of her lungs was decreased with crackles, but she did not have lymphadenopathy or weight loss. Furthermore, we could hear stridor in the trachea and neck.

Laboratory examinations

Routine blood tests showed an elevated white blood cell count (14.70 × 10^9 cells/L; range, 3.5-9.5 × 10^9 cells/L), absolute neutrophil count (11.36 × 10^9 cells/L; range, 1.8-6.3 × 10^9 cells/L), and neutrophil percentage (77.3%; range, 40%-75%); the serum potassium level was found to be decreased in the blood biochemistry results (2.78 mmol/L; range, 3.5-5.5 mmol/L). The tumor markers were normal. The arterial

blood gas test suggested respiratory acidosis combined with metabolic alkalosis.

Imaging examinations

Pneumonia was detected from the chest X-ray, with no other abnormalities. A computed tomographic (CT) scan of the chest showed a sign of pulmonary infection, and computed tomographic virtual bronchoscopy (CTVB) showed a well-defined, soft-tissue density lesion measuring 2.4 cm × 2.1 cm in the lower trachea, located 2 cm above the carina (Figure 1). Fiberoptic bronchoscopy revealed that the surface of the mass was smooth and vasodilatory, and nearly 90% of the tracheal lumen was obstructed, so the bronchoscope failed to pass through (Figure 2).

Pathological examination

Histopathological analysis revealed that the tumor was composed of epithelial and myxoid mesenchymal elements and was characterized by the presence of ductal structures that appeared to contain double-layered cells in a mucoid or hyaline stroma. Notably, there was no sign of necrosis or mitosis (Figure 3). Immunohistochemically, the tumor cells did not express thyroid transcription factor-1 and cytokeratin 7 (CK 7), but were positive for CK, CK 5/6, p63, and the S-100 protein, with low expression of Ki-67 (10%). Moreover, the basement membrane was immunoreactive for AB/ para-aminosailcylic acid. After immunohistochemical staining, the definite diagnosis was determined to be PA of the trachea.

FINAL DIAGNOSIS

The patient was finally diagnosed with PA of the trachea.

TREATMENT

Considering that the patient's vital signs were stable, intratracheal tumor resection was performed by electron bronchoscopy under conscious sedation induced using intravenous midazolam. Finally, tumor tissues were excised with an electro-surgical snare and cryotherapy. Then, the edges and base of the mucosal defect were treated with argon plasma coagulation (APC) to enhance tumor clearance. There was no significant bleeding or perforation from the wound (Figure 2). After resection, the tracheal lumen was completely unobstructed, and there were no new organisms.

OUTCOME AND FOLLOW-UP

The patient's wheezing symptoms were remarkably relieved after the operation, but cough and expectoration remained. Regarding the sign of pulmonary infection from the chest CT, the patient was discharged 9 d after anti-infection treatment and remained asymptomatic at the 3-mo follow-up.

DISCUSSION

PA originating from the trachea is rare. According to Fitchett *et al*[11], it accounts for 1% of lung carcinomas and between 2% to 9% of all cases of PA. This type of PA consists of myoepithelial cells mixed with neoplastic ducts and stroma. The demographics and presenting characteristics of the 29 cases are shown in Table 1. Likewise, the major clinical features of the patients are listed in Table 2. According to the review, no gender predominance was found. The age of the patients ranged from 8 to 83 years, with a median age of 48 years, and there were four minors. More than half of these tumors were located in the lower or upper trachea; however, two cases originated from the airway and grew outward into the thyroid or mediastinum. Although a few patients presented with hemoptysis, the most common symptoms were cough, stridor, dyspnea, and wheezing, depending on the site and degree of airway obstruction. The patient in this case had a 2-year history of dyspnea upon exertion and chronic cough with wheezing before being properly diagnosed with PA of the trachea. The median clinical course was 5.5 mo, and the longest course

Table 1 Summary of presenting characteristics of tracheobronchial pleomorphic adenoma reported in the English medical literature

Ref.	Age	Sex	Clinical presentation	Course (mo)	Tumor site	Tumor size (cm)	Immunohistochemical staining	Treatment	Comorbidities	Complications	Clinical follow-up period (mo)
Heifetz *et al*[18], 1992	15	M	Asthma, wheezing, and dyspnea	12	Upper trachea (level of the fourth ring)	2.5 × 2.5 × 2.5	+: CK AE1/3, S-100, actin, vimentin, EMA, GFAP	CO2 laser bronchoscopy	No	No	Alive with no evidence of recurrence (6)
Basaklar *et al*[19], 1994	11	F	Nonproductive harsh cough, high fever, nausea, vomiting, and night sweats	1.5	Right upper lobe bronchus	2	Not available	Surgical resection	Atelectasis, multiple mediastinal and peribronchial lymphadenopathies	No	Not available
Sweeney *et al*[20], 1996	27	M	Incidental (asymptomatic)	Not available	Right lower lobe bronchus	3 × 5	+: CK, EMA, S 100, SMA	A lower lobectomy	No	No	Not available
Paik *et al*[21], 1996	50	M	Mild dyspnea upon exertion	3	Mid trachea (4 cm above the carina)	2 × 2	Not available	Right thoracotomy with segmental resection and end-to-end anastomosis	No	No	Alive with no evidence of recurrence (18 d)
Bizal *et al*[22], 1997	27	M	Dyspnea upon exertion and intermittent wheezing	12	Lower trachea (2 cm above the carina)	2.5	Not available	Surgical resection and primary anastonosis performed through right thoracotomy	No	No	Alive with no evidence of recurrence (6)
Paik *et al*[23], 1997	48	F	Dyspnea upon exertion and productive cough with wheezing	3	Lower trachea	1.5 × 1.2	+: Vimentin, CK, S-100, GFAP, SMA	Tracheal wedge resection	No	No	Not available
Pomp *et al*[24], 1998	79	F	Increasing stridor, dyspnea and a dry cough	2	Upper trachea (level of fifth ring)	2	Not available	Radiotherapy, excision through rigid bronchoscopy	No	Recurrent PA of the trachea	Not available
Pomp *et al*[24], 1998	58	F	Increasing dyspnea and stridor	6	Upper trachea (below the larynx)	90% occlusion	Not available	Excision *via* tracheotomy	No	No	Alive with no evidence of recurrence (12)
Kim *et al*[25], 2000	15	M	Asthma, dyspnea and stridor	5	Upper trachea	1.5	Not available	Segmental tracheal resection and end-to-end anastomosis	No	No	Alive with no evidence of recurrence (12)
Baghai-Wadji *et al*[7], 2006	8	M	Asthma, fever, productive cough, severe wheezing, and respiratory distress	10 d	Lower trachea	90% occlusion	+: Chromogranin, NSE, CK	Surgical resection and tracheal reconstruction (pericardial patch graft)	Pneumonia	No	Alive with no evidence of recurrence (6)
Aribas *et al*[8], 2007	42	F	Asthma, severe dyspnea	2 yr	Lower trachea	2 × 2	+: Vimentin, GFAP, S-100	Segmental tracheal resection and end-to-end anastomosis	No	Tracheal stenosis	Alive with no evidence of recurrence (5 yr)

Ashwaq *et al*[26], 2007	37	M	Spontaneous hemoptysis	8	Mid trachea	2 × 2	Not available	Excision with cold instrument *via* suspension laryngoscopy	No	No	Alive with no evidence of recurrence (3)
Matsubara *et al*[27], 2008	71	M	Incidental (asymptomatic)	Not available	Left main bronchus	Not available	+: polyclonal anti-S-100, anti-GFAP	Endoscopic resection with electrosurgical snaring and APC	No	No	Alive with no evidence of recurrence (6)
Fitchett *et al*[11], 2008	65	M	Hoarse barking cough	5	Right main bronchus	1.3	Not available	Endoscopic resection with diathermy snare	No	No	Not available
Kamiyoshihara *et al*[28], 2009	34	F	Dyspnea upon exertion	3	Left main bronchus	1.2 × 1.1	Not available	Surgical resection with wedge bronchiectomy	No	No	Alive with no evidence of recurrence (11)
Tanaka *et al*[13], 2010	57	F	A neck mass	10 yr	Right lobe of the thyroid (originating from the trachea)	3.25 × 2.09	+: SMA, 34bE12; -: P53 and ki67	Surgical resection and direct anastomosis	No	No	Not available
Kajikawa *et al*[9], 2010	55	M	Asthma, dyspnea with wheezing	2 yr	Lower trachea	Not available	Not available	Endoscopic resection with APC, electrocautery and rigid bronchoscopic coring	No	No	Alive with no evidence of recurrence (7)
Lin *et al*[29], 2011	36	F	Bronchial asthma, worsening shortness of breath	6	Lower trachea(3 cm above the carina)	2 × 2 × 2	Not available	Segmental tracheal resection and anastomosis	Allergic rhinitis	No	Not available
Goto *et al*[30], 2011	71	M	Progressive dyspnea	Not available	Left main bronchus	2.5 × 2	+: CK AE1/3, SMA	Endoscopic resection with electrosurgical snaring	Chronic obstructive pulmonary disease, squamous cell; carcinoma (pT2N0M0, stage IB)	No	Alive with no evidence of recurrence (2)
Solak *et al*[15], 2012	46	F	Severe dyspnea	12	Upper trachea	3 × 2	Not available	Collar incision with partial sternotomy and end-to-end anastomosis	No	No	Alive with no evidence of recurrence (1)
Park *et al*[16], 2013	59	M	Dyspnea upon exertion	3	Mid trachea	2 × 2	+: CK, CK 19, EMA, S100, p63	Right thoracotomy with segmental resection and end-to-end anastomosis	Active pulmonary tuberculosis	No	Alive with no evidence of recurrence (5 yr)
Lee *et al*[31], 2014	54	F	Blunt chest pain upon bending forward	2 wk	Posterior mediastinum (originating from the left main bronchus)	6.0 × 4.5 × 2.5	+: P63 and SMA	Video-assisted thoracic surgery	No	No	Alive with no evidence of recurrence (2 yr)
Casillas-Enríquez *et al*[32], 2014	33	F	Productive cough, wheezing, and occasional hemoptysis	4 yr	Upper trachea	80% occlusion	Not available	Endoscopic resection with APC	No	No	Alive with no evidence of recurrence (8)
Sim *et al*[33], 2014	32	F	Dyspnea upon exertion and chronic cough with wheezing	8	Lower trachea	1.8 × 1.6	Not available	Endoscopic resection with rigid forceps and APC	Situs inversus	No	Alive with no evidence of recurrence (1)

Zhu *et al*[3], 2018	38	F	Progressive shortness of breath	5 yr	Right main bronchus	1.42 × 0.96	Not available	Endoscopic resection with electrosurgical snare and APC	No	No	Alive with no evidence of recurrence (3)
Kim *et al*[4], 2018	49	M	Exacerbation of dyspnea upon exertion, cough and sputum	3	Lower trachea	1.5 × 1.3 × 1.3	+: CK 5/6, CK, p53	Right thoracotomy with segmental resection and anastomosis with tracheobronchoplasty	Active pulmonary tuberculoma	No	Alive with no evidence of recurrence (3)
David *et al*[5], 2020	83	F	Worsening shortness of breath and waking up with blood in her oropharynx	1	Upper trachea (3.0 cm below the vocal fold edge)	1.6 × 1.3	+: P63, SMA; -: Chromogranin, synaptophysin	Endoscopic excision with fiber-based CO2 laser and rigid bronchoscope	Hypertension, rheumatoid arthritis	No	Not available
Takahashi *et al*[6], 2019	51	F	Asthma, cough and wheezing at night	2	Upper trachea (periphery 30 mm from the glottis)	1.5	Not available	Endoscopic resection with electrosurgical snaring and forceps	No	No	Alive with no evidence of recurrence (30)
Our case	49	F	Dyspnea upon exertion and chronic cough with wheezing	2 yr	Lower trachea	2.4 × 2.1	+ □CK, CK 5/6, p63, S-100, Ki-67 (10%); - □TTF-1, CK 7	Endoscopic resection electrosurgical snare, cryotherapy and APC	No	No	Alive with no evidence of recurrence (3)

CK: Cytokeratin; EMA: Epithelial membrane antigen; GFAP: Glial fibrillary acidic protein; SMA: Smooth muscle actin; NSE: Neuron-specific enolase; APC: Argon plasma coagulation; TTF-1: Thyroid transcription factor-1; M: Male; F: Female; CK 7: Cytokeratin 7.

was 10 years, which may reflect the benign nature of the tumor. In addition, it results in low recurrence rates at follow-ups.

Tracheal tumors are difficult to identify in chest radiographs. Moreover, patients initially present with non-alarming symptoms mimicking asthma[11]. The patient in this case was previously misdiagnosed with asthma and treated with inhaled glucocorticoids for 2 mo. Therefore, chest CT and bronchoscopy play a critical role in making early and proper diagnoses. CTVB involves the three-dimensional reconstruction of high-resolution helical CT images of the tracheobronchial tree, which can facilitate the analysis of bronchial lesions beyond the limits of bronchoscopy and the assessment of airway patency distal to high-grade obstructions[12]. However, CTVB cannot be used to identify the nature of a lesion, while bronchoscopy can be used to complete this by biopsy.

Histologically, PA is also known as a "mixed tumor", which describes its pleomorphic appearance rather than its dual origin from epithelial and mesenchymal components. The stroma may be mucoid, myxoid, cartilaginous, or hyaline. Approximately 6% of tumors have the potential to transform into carcinoma ex pleomorphic adenoma[10]. When it presents with atypical cells, an abnormal chromatin pattern, and necrosis, the diagnosis of carcinoma ex pleomorphic adenoma is made. Regarding immunohistochemistry findings, the tumor shows positive staining for creatine kinase, p63, S-100 protein, epithelial membrane antigen, and glial fibrillary acidic protein. S-100 protein and glial fibrillary acidic protein may be helpful markers in differentiating PA and adenoid cystic carcinoma[13]. In addition, the patient in our

Table 2 Outline of major features characterizing presentation of 29 cases of tracheobronchial pleomorphic adenoma

Variable	*n* (%) or median (IQR)
Sex	
Female	16 (55.17)
Male	13 (44.83)
Age, yr	
Median (range)	48 (8-83)
Symptoms	
Asymptomatic	2 (6.90)
Respiratory symptoms (wheezing, dyspnea, cough, stridor, hemoptysis)	24 (82.76)
Fever	2 (6.90)
Gastrointestinal symptoms (vomiting, diarrhea)	1 (3.45)
Night sweats	1 (3.45)
Chest pain	1 (3.45)
Neck mass	1 (3.45)
Clinical course	
Median (range)	5.5 m (10 d-10 y)
Location	
Upper trachea	8 (27.59)
Mid trachea	3 (10.34)
Lower trachea	9 (31.03)
Bronchus	7 (24.14)
Thyroid	1 (3.45)
Posterior mediastinum	1 (3.45)
Size (largest diameter), cm	
Median (range)	2 (1.2-6)
Recurrence	1 (3.45)

IQR: Interquartile range.

study had a Ki-67 index of 10%. This marker is widely known as a proliferative marker, and numerous studies have shown a positive correlation between Ki-67 expression and the proliferative cell fraction in tumors[14].

Given the rarity of tracheal PA, no standards for management have been established, but it is clear that the main goal is to remove the lesion and restore airway patency. Surgical resection and airway anastomosis have traditionally been applied in many studies[4,15,16]. Compared with surgery, endoscopic resection is less traumatic and allows a faster recovery after the operation. Endobronchial intervention using a rigid and flexible bronchoscope is widely performed in cases of airway stenosis. In our case, we successfully applied bronchoscopic interventional therapy to remove the tumor, such as electrosurgical snare, cryotherapy and argon plasma coagulation. Due to its rarity, its biological behavior and clinical course have not been well described. One case of tracheal PA was reported to be recurrent in 2020 after surgical resection and end-to-end anastomosis were performed 10 years previously[17]. Therefore, long-term follow-ups are essential for patients. According to the medical literature, there is no clearly recommended follow-up period or interval, of which the longest follow-up period is 5 years without recurrence[8]. We will follow this patient by periodic chest CT and flexible bronchoscopy at least 10 years after the tumor resection.

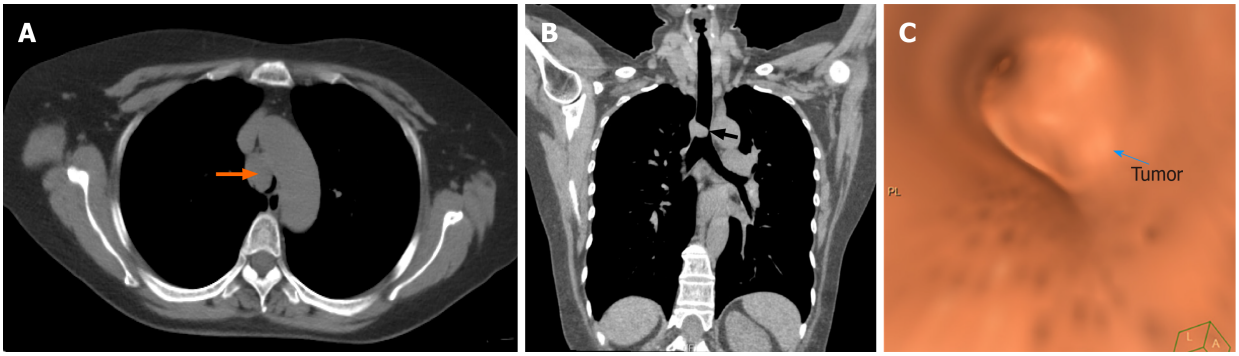

Figure 1 Computed tomographic presentation of the patient. A: Mediastinal computed tomographic scan of the chest showed a 2.4 cm × 2.1 cm homogenous well-defined, dense soft tissue lesion in the left lateral inner wall of the trachea (orange arrows); B: Computed tomographic scan with multiplanar reconstruction showed a round lesion in the lower trachea (black arrow); C: A tumor in the inner trachea observed by computed tomographic virtual bronchoscopy (blue arrow).

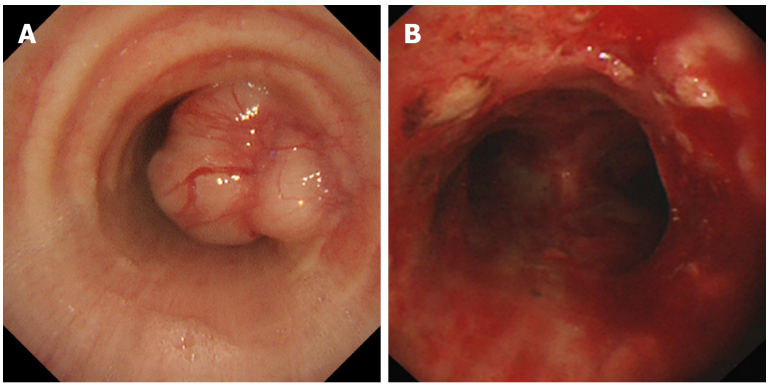

Figure 2 Bronchoscopic findings. A: A polypoid and vasodilatory mass originated from the right side of the lower trachea; B: After endoscopic resection, the tumor was removed almost completely, and the airway patency was restored.

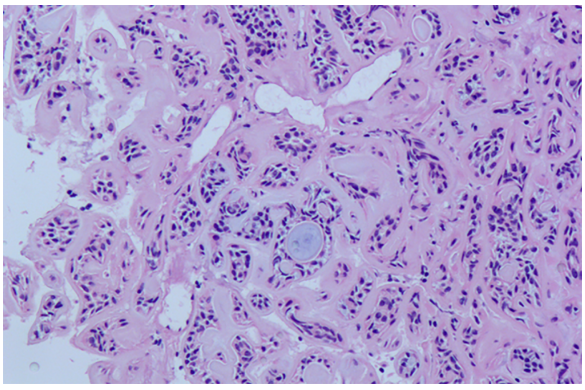

Figure 3 Pathological presentation of the patient. The tumor was composed of epithelial and myxoid mesenchymal elements and characterized by the presence of ductal structures that appeared to contain double-layered cells in a mucoid or hyaline stroma. No signs of necrosis or mitosis were observed (hematoxylin-eosin staining, × 100).

CONCLUSION

Overall, we summarize the clinical presentation, clinical course, treatment, and prognosis of tracheobronchial PA according to the literature over the last 30 years[18-33]. PA of the trachea is extremely rare, and patients initially present with non-specific symptoms mimicking asthma. Chest CT and bronchoscopy play a critical role in making an early diagnosis, whereas a definite diagnosis is made on the basis of histopathological and immunohistochemistry features. Although surgical resection is traditionally performed, this article supports the notion that bronchoscopic

interventions for PA of the trachea are viable treatment options.

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