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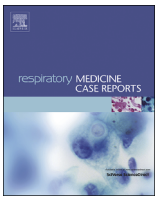

Case Report

Long-term follow-up of tracheal pleomorphic adenoma: A case report

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ABSTRACT

Pleomorphic adenomas occur primarily in the salivary glands, while the primary of the trachea is relatively rare. Depending on their location and size, they may elicit symptoms reminiscent of asthma and asphyxia. We performed a rigid bronchoscopic resection with a radiofrequency snare of a primary pleomorphic adenoma of the trachea with severe airway narrowing. In this case, the positive resection margins raised concerns about malignant transformation and local recurrence, but no recurrence has been observed seven years postoperatively. The low expression of Ki-67 on immunohistological examination may be one of the reasons for the absence of recurrence.

1. Introduction

This case report highlights a patient with primary tracheal pleomorphic adenoma who underwent successful excision via snare resection under rigid bronchoscopy. Despite positive margins, the patient has remained recurrence-free for seven years, suggesting a potential association with the tumor's Ki-67 low expression.

2. Case presentation

A 53-year-old woman was diagnosed with an asthma attack by her primary care physician and was treated with medication. However, her dyspnea worsened, so she went to the emergency department, where a chest Computed tomography (CT) (Fig. 1A) revealed severe airway narrowing due to a mass occupying approximately 90 % of the tracheal lumen.

On admission, the patient did not have a fever and had obvious wheezing on chest auscultation. A bronchoscopy was performed on the day of admission, and the flexible bronchoscope showed a smooth surface (Fig. 2A).

During bronchoscopy, the cough reflex became stronger, and ventilation became difficult, so the patient was endotracheally intubated and managed in the intensive care unit. The patient had severe stenosis, but the tumor was considered benign, and the decision was made to perform under rigid bronchoscopic resection. Assuming the possibility of intraoperative ventilatory failure, the cardiovascular surgeon was consulted to prepare for extracorporeal membrane oxygenation (ECMO), and both patient's groins were exposed, and the femoral arteriovenous veins were marked. The patient was sedated to the extent that spontaneous breathing remained,

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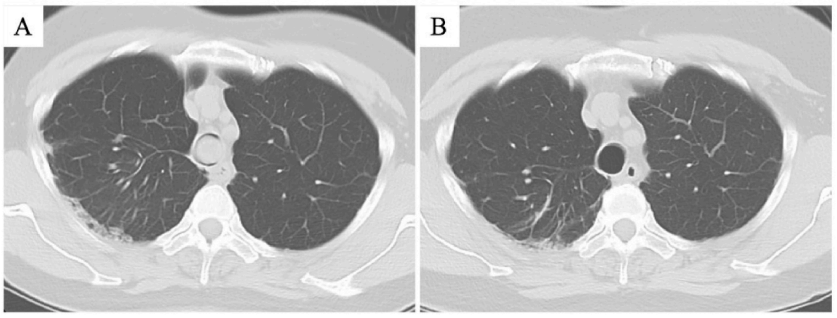

Fig. 1. Computed tomography (CT) shows severe tracheal obstruction due to a tumor on admission (A). CT showing no recurrence after 7 years post-resection (B).

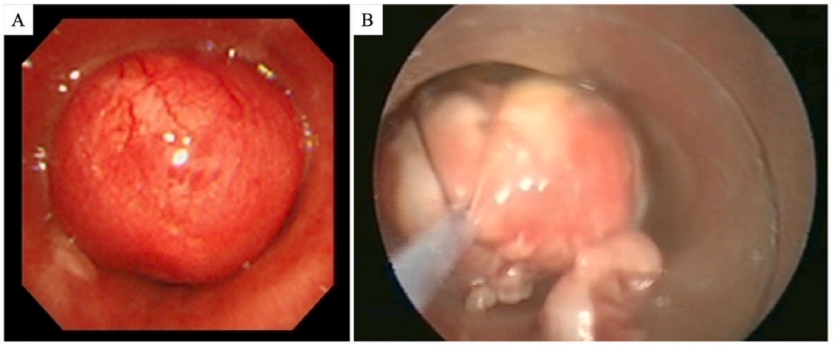

Fig. 2. Flexible bronchoscopy findings at admission showed severe tumor obstruction of the main bronchus (A). Retrieved with a basket forceps after snare resection under rigid bronchoscopy (B).

and after resection using a rigid bronchoscope with a high-frequency snare, the patient was removed with basket forceps (Video 1, Fig. 2B).

Supplementary video related to this article can be found at https://doi.org/10.1016/j.rmcr.2024.102073

The resection was completed without the use of ECMO. Bronchoscopy approximately 10 months later showed no recurrence at the tumor margins. Thereafter, during 7 years of outpatient follow-up, there was no recurrence (Fig. 1B).

Pathologic examination revealed localized and cordate proliferation of round, cuboidal, and polygonal cells with vitrified stroma within the tumor. Fat cells were extensively present, and a diagnosis of pleomorphic adenoma was made. In addition, there were positive margins and little or no Ki-67 labeling.

3. Discussion

Primary tracheal tumors are rare, and their frequency is reported to be approximately 1 % of lung tumors [1]. Furthermore, approximately half of the pleomorphic adenomas tend to occur in the salivary glands, especially the parotid gland, making the primary pleomorphic adenoma of the trachea a very rare entity. Literature reports of primary pleomorphic adenomas of the trachea show respiratory symptoms in many cases (Table 1). There have been reports of asthma treatment before the diagnosis of primary tracheobronchial pleomorphic adenoma, especially because of the difficulty in differential diagnosis from bronchial asthma [2–4]. Some cases of tracheal pleomorphic adenoma present with severe airway narrowing in more than 80 % of cases with asthma-like symptoms such as cough and dyspnea. However, the tumor may be asymptomatic because its primary location was in the left main bronchus, and it may be discovered incidentally, e.g., during the examination of other diseases [5,6]. A case with complete occlusion of the right main bronchus had only symptoms of coughing [7], but the case with complete occlusion just above the carina was accompanied by impaired consciousness.3 Primary tracheal tumors may be asymptomatic, with symptoms varying according to the site of origin, tumor size, and stenosis rate. In this study, the patient had been treated for bronchial asthma since his 30s, and his cough was diagnosed as an asthma attack. However, the patient was referred to our clinic because of dyspnea and poor response to drug therapy. Wheezing episodes refractory to drug therapy require chest CT evaluation to determine the possibility of organic stenosis. Airway stenosis may be present, and resection is the mainstay of treatment. If severe stenosis is present at the initial presentation, airway clearance is mandatory. In this case, the patient developed a severe cough following stimulation by flexible bronchoscopy performed during an initial visit to another department. Although he was in ventilatory failure, he was immediately ventilated by deepening the tracheal tube. Flexible bronchoscopy in patients with severe airway stenosis should be performed with caution. Preparation and use of ECMO should be considered to resolve ventilatory insufficiency [3], but in many cases, including ours, resection was possible without ECMO [2,4–9]. Flexible bronchoscopic snare resection [5,6], APC [2,6,8], and surgical resection have been reported for tumor resection [3,4,7,9]. APC is considered useful for cauterization and hemostasis because of its shallow penetration into deep tissue. It may be use-

Table 1
Literature review of tracheal pleomorphic adenoma.

Case	Author (year)	Age	Sex	Symptoms at diagnosis	History of BA	Tumor locate	Tumor size	Stenosis rate	Treatment	ECMO	Follow-up time/recurrence
1	Sim et al. (2014)	32	F	Dyspnea	No	Trachea	18mm	90 %	APC under rigid BFS	No	1 month/No
2	Casillas-Enriquez et al. (2014)	33	F	Wheezing, cough, hemorrhage	Yes	Trachea	NA	80 %	APC under rigid BFS	No	8 months/No
3	Silva et al. (2023)	66	M	None	No	Trachea	12mm	NA	Sleeve resection	No	NA
4	Zhong et al. (2020)	10	M	Cough	No	RMB	13mm	100 %	Right upper lobectomy	No	1 month/No
5	Inomata et al. (2023)	69	F	Dyspnea, LOC	Yes	Above the carina	15mm	100 %	Carina resection	Yes	12 months/No
6	Solak et al. (2012)	46	F	Asthma attack	Yes	Trachea (Inside and outside of trachea)	30mm	90 %	Sleeve resection	No	1 month/No
7	Goto et al. (2011)	71	M	None (at diagnosis with LC)	No	LMB	25mm	60 %	Snare resection under flexible BFS	No	Following/No
8	Matsubara et al. (2008)	71	M	None	No	LMB	NA	NA	Snare resection and APC under flexible BFS	No	6 months/No
9	Current case	54	F	Dyspnea	Yes	Trachea	19mm	90 %	Snare resection under rigid BFS	No	7 years/No

BA: Bronchial asthma, ECMO: Extracorporeal membrane oxygenation, NA: Not available, APC: Argon plasma coagulation, BFS: Bronchofiberscopy, RMB: Right main bronchus, LOC: Loss of consciousness, LMB: Left main bronchus, LC: Lung cancer.

ful when the tumor has no stalk and cannot be snared. We used a rigid bronchoscope for resection because of concerns about ventilation failure. Another advantage of rigid bronchoscopy is its ability to resect tumors by debulking. Pleomorphic adenomas are benign tumors that have a 3–15 % risk of malignant transformation in the distant postoperative period [10], and capsular injury during resection also increases the risk of recurrence [11,12]. Ki-67 is a protein that is released during cell division and is a marker of tumor growth. It is present during the active phase of the cell cycle and absent from resting cells [13]. Reports correlating Ki-67 expression with the prognosis of pleomorphic adenomas emphasize the importance of paying close attention to this biomarker [11,12]. The fact that patients remained recurrence-free for seven years postoperatively despite positive resection margins in our case, while most reports have a follow-up period of less than 1 year postoperatively, can be attributed to low Ki-67 expression.

4. Conclusion

Primary pleomorphic adenoma of the trachea is a rare benign disease, but Ki-67 may be useful as an indicator of recurrence and possible malignant transformation.

Consent for publication

Written informed consent for publication was obtained from the patient discussed in this article.

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CRediT authorship contribution statement

Naoya Ishibashi: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. **Toshiharu Tabata:** Writing – review & editing. **Ryo Nonomura:** Writing – review & editing. **Yutaka Oshima:** Writing – review & editing. **Takanobu Sasaki:** Writing – review & editing. **Hideki Mitomo:** Writing – review & editing. **Takafumi Sugawara:** Writing – review & editing. **Motoyasu Sagawa:** Writing – review & editing. **Nobuyuki Sato:** Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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