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Case report

Pneumothorax as a rare presentation of bronchial schwannoma

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ABSTRACT

Tracheobronchial schwannomas are rare diseases. Common signs and symptoms of this tumor include cough, wheezing, and dyspnea. In contrast, pneumothorax is an exceptional presentation. This study reports the first case of bronchial schwannoma presenting with pneumothorax. A 79-year-old woman was diagnosed with pneumothorax by chest radiography. Chest computed tomography unexpectedly revealed a tumor occluding the right main bronchus. Following the pathological diagnosis of bronchial schwannoma, the patient underwent thoracoscopic tumor enucleation. The airway lumens are consequently secured postoperatively. We reviewed the literature and discussed the mechanisms and treatment options for bronchial benign tumor-associated pneumothorax. Pneumothorax should be aware of a rare presentation of non-malignant tracheobronchial tumors.

1. Introduction

Tracheobronchial tumors are mostly malignant or intermediate malignancies, and benign tumors, especially schwannomas, are rare. Schwannomas are tumors originating from the peripheral nerve sheath anywhere in the whole body, and rarely affect the tracheobronchial tree. The presentations of tracheobronchial tumor are non-specific, depending on the size and location, while pneumothorax exceptionally reveals bronchial tumors. To the best of our knowledge, we describe the first case of bronchial schwannoma presenting with pneumothorax.

2. Case presentation

A 79-year-old woman was referred to our department after being diagnosed with right-sided pneumothorax by chest radiography during a routine health check-up. The patient received acupuncture treatment 6 weeks prior; however, remained asymptomatic during and after the procedure. Her vital signs were normal and routine laboratory tests revealed unremarkable findings. The respiratory sounds were attenuated in the right chest. Chest radiography showed a moderate degree of right-sided pneumothorax and mediastinal shift to the ipsilateral side (Fig. 1A). Following tube thoracostomy, computed tomography (CT) of the chest revealed right-sided pneumothorax, ipsilateral mediastinal shift, and a small amount of pleural effusion. In addition, a tumor (maximum diameter of 25

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mm) was present in the posterior direction of the right main bronchus, almost completely obstructing the airway lumen (Fig. 1B and C). It should be notable that the post-obstructive lung volume was reduced, and bronchiectasis was apparent on CT, suggesting that the fixed bronchial stenosis by the tumor disrupted air entry to the post-obstructed lungs and increased negative pressure in the pleural space. A retrospective review of the chest CT performed 3 years ago revealed a smaller tumor on the posterior wall of the right main bronchus with the bronchial lumen being spared, and the lung parenchyma appeared to be normal without a loss of volume or bronchiectasis (Fig. 1D and E). Bronchoscopy revealed an extraluminal bronchial tumor that almost completely obstructed the right main bronchus (Fig. 1F). Taken together, the slowly progressive tumor on the bronchial wall obstructed the right main bronchus, which subsequently reduced the post-obstructed lung volumes and was presumably related to the pneumothorax.

The patient underwent percutaneous core needle biopsy, and the pathological diagnosis of bronchial schwannoma was made preoperatively. Since bronchial obstruction could presumably cause insufficient lung expansion and sustained air leakage through the chest tube, surgical resection of the tumor was scheduled. During the operation, the tumor was tightly adhered to the bronchial wall and could hardly be removed without bronchial resection. We finally performed enucleation without bronchial resection, instead of complete resection of the encapsulated tumor, as it is less invasive for benign tumors in elderly patients. No leaking hole was found on the visceral pleura during the operation. The pathological examination of the surgical specimen showed a white yellowish solid tumor 25 mm in diameter, which was surrounded by a fibrous capsule (Fig. 2A). Microscopic examination showed dense proliferation of spindle-shaped tumor cells with elongated nuclei in hematoxylin-eosin staining (Fig. 2B). These cells were positive for the S-100 protein (Fig. 2C) but were almost negative for Ki-67 (not shown) in immunostaining. These microscopic findings confirmed a pathological diagnosis of schwannoma identical to that of the preoperative biopsy specimen. The pneumothorax improved postoperatively, and the patient was discharged from the hospital. The secured airway lumen in the right main bronchus was confirmed two months after surgery (Fig. 3A). The patient's respiratory symptoms were absent, and no substantial tumor regrowth was observed for 3 years after the surgery; lung volumes were not restored. (Fig. 3B and C).

3. Discussion

Tracheobronchial tumors are mostly malignant, and benign tumors account for 4.7% of cases [1]. Among these, schwannomas are extremely rare and have been reported in four out of 185 (2.2%) patients with benign tracheobronchial tumors [1]. Schwannoma is a tumor that occurs from the peripheral nerve sheath anywhere in the whole body, and rarely affects the autonomous nerves in the bronchial wall. Tracheobronchial schwannoma affects any site of tracheobronchial trees intra- or extra-luminally and grows gradually. Common signs and symptoms of this tumor include cough, wheezing, and dyspnea. Less common symptoms include hemoptysis, hoarseness, and chest pain, depending on the size, location, and extent of airway narrowing by the tumor. In contrast, pneumothorax is an exceptional presentation of tracheobronchial tumors, regardless of the pathological type. We comprehensively reviewed the literature and identified as few as two cases of benign bronchial tumors initially presenting with pneumothorax, one chondroma [2] and one stromal tumor [3]. We additionally found tracheobronchial carcinoid cases, intermediate malignant tumors of neuroendocrine cells, presenting with pneumothorax [4–8]. Two cases [7,8] were excluded from further discussion because of difficult accession to the literature. The three remaining cases were typical carcinoids that were an entity of slow progressive tumors. Table 1 summarizes the clinical characteristics of the cases. Major symptoms at the time of diagnosis included chest pain, cough, and breathlessness, while the

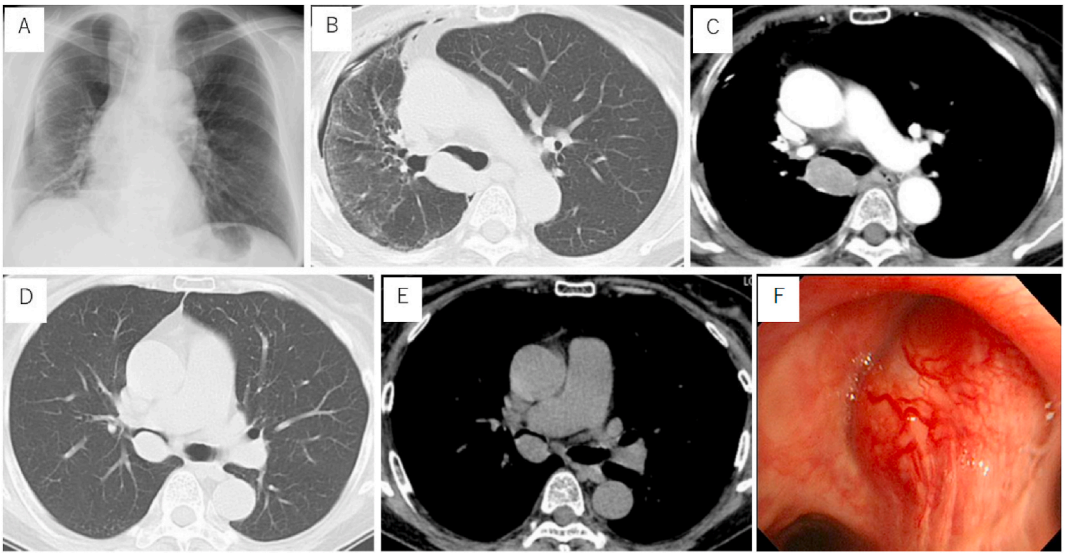

Fig. 1. Chest radiography showed a moderate degree of right-sided pneumothorax and a mediastinal shift to the ipsilateral side (A). Contrast medium-enhanced chest computed tomography (CT) revealed a tumor located in the posterior direction of the right main bronchus, almost completely obstructing the airway lumen (B, C). A smaller tumor was located on the posterior wall of the right main bronchus as seen on CT obtained 3 years ago (D, E). Bronchoscopy revealed an extraluminal tumor that almost completely obstructed the right main bronchus (F).

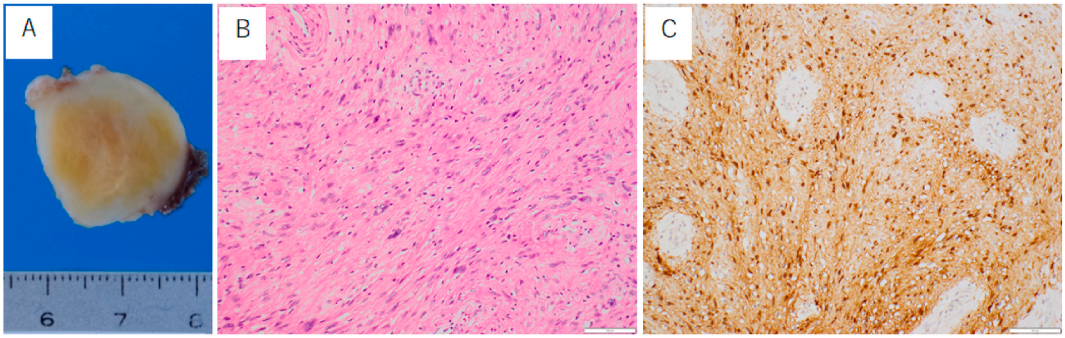

Fig. 2. Macroscopic findings show a white yellowish tumor with fibrous capsule formation (A). The tumor is microscopically composed of a dense proliferation of spindle-shaped cells with elongated nuclei in hematoxylin and eosin staining (B). The tumor cells were positive for the S-100 protein in immunostaining (C). Scale bars = 100 μm.

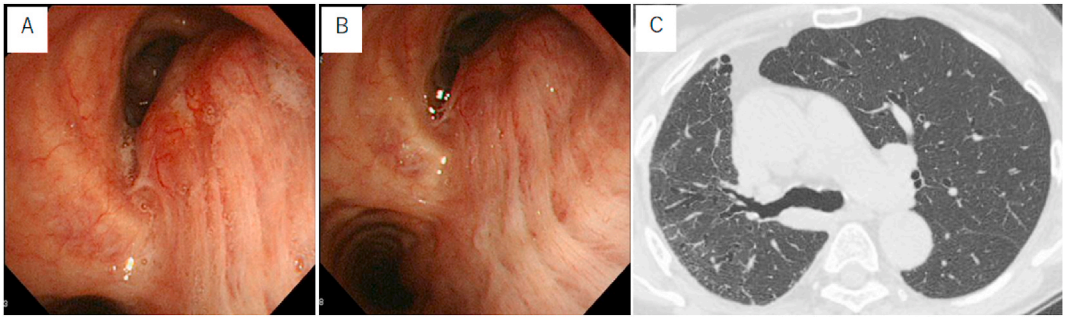

Fig. 3. Bronchoscopy showed a secured airway lumen in the right main bronchus two months after the surgery (A). Substantial tumor regrowth of the right main bronchus was absent on bronchoscopy (B) and chest computed tomography (C) for 3 years postoperatively.

patient in the present case was asymptomatic and the disease was found on chest radiography as a routine health check-up. The tumors were located on the main bronchus in four cases and on the lobar bronchus in two cases. Intraluminal locations were common.

The mechanisms underlying bronchial tumor-associated pneumothorax are not well understood. Rupture of metastasizing tumors in the pleural or subpleural zone is the most frequent cause of pneumothorax in malignant tumors but is rare in benign tumors. In contrast, benign non-metastatic tumors are potentially associated with pneumothorax in two different ways. One mechanism is a check-valve formation and overinflation of post-obstructive lungs, leading to a rupture of air cysts in the subpleural space [5]. The mechanism might be associated with the high flexibility of the intraluminal polypoid tumors as typical carcinoid cases [4–6]. Another mechanism was applicable in previous [2,3] and present cases, in those the transluminal tumor or long-segment bronchial cast caused fixed airway stenosis and disrupted air entry to the post-obstructive lungs. This condition reduced the post-obstructive lung volume and theoretically increased negative pressure in the pleural space. Finally, elevated transpulmonary pressure was potent to harm the visceral pleura or draw gas into the pleural space from the ambient tissue despite the intact pleura (pneumothorax ex vacuo) [9]. In

Table 1
Reported cases of non-malignant bronchial tumors presenting with pneumothorax.

Age, sex	Symptom	Pathology	Location		Size (mm)	Treatment	Ref.
20, F	cough chest pain breathlessness	chondroma	right main bronchus	combined	–	tumor enucleation	2
34, M	fever sputum breathlessness	stromal tumor	left lower bronchus	intraluminal	–	left lower lobe resection removal of trachea-bronchial cast	3
30, F	chest pain cough hemoptysis	typical carcinoid	left main bronchus	intraluminal	20	sleeve resection left upper lobe lobectomy	4
65, M	chest pain breathlessness	typical carcinoid	left upper bronchus	intraluminal	–	sleeve resection	5
18, M	fever chest pain breathlessness	typical carcinoid	left main bronchus	intraluminal	16	laser photocoagulation surgical resection (detailed unknown)	6
79, F	none	schwannoma	right main bronchus	combined	25	tumor enucleation	present

addition, a case-specific mechanism should also be considered in conjunction with acupuncture in the present case. Since no additional images were available between the acupuncture and the hospital visit, we could not conclude if the acupuncture caused pneumothorax. In the case of acupuncture-associated pneumothorax, the bronchial tumor is complicit in progressing that via disrupting air entry to the collapsed lungs.

The management of tracheobronchial tumors should differ depending on the pathological type, tumor size, location, symptoms, and patient's condition. The dominance of the extraluminal component is an important determinant. All three cases of typical carcinoid accompanying pneumothorax required bronchial sleeve resection for complete resection (Table 1). In contrast, the treatment strategies for tracheobronchial benign tumors are less invasive. Kasahara et al. proposed a classification of the tracheobronchial schwannoma based on tumor location: central-intraluminal, central-combined (both intra- and extra-luminal), and peripheral types [10]. Several cases with the central-intraluminal type showed favorable outcomes in intraluminal bronchoscopic procedures [10]. On the other hand, the present case was applied to the central-combined type, for which surgical resection was recommended. Due to the extra-luminal predisposition of the tumor in the present case, we enucleated the tumor without bronchial resection as it was less invasive for elderly patients. Consequently, the patient was free from respiratory symptoms for a long period without substantial tumor regrowth.

4. Conclusion

We report a case of bronchial schwannoma presenting with pneumothorax. Pneumothorax should be aware of a rare presentation of non-malignant tracheobronchial tumor. Post-obstructive lung volume was a key finding to assume the mechanisms of pneumothorax. The appropriate surgical procedure helped in the successful management of the tracheal tumor with pneumothorax.

Consent

Written consent was obtained from the patient for publication of this case report and for a use of accompanying images.

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Submission declaration and verification

The present study was not published or is not currently submitted to any other journal.

Authors' contribution

All of the authors ensure to task force for preparing the manuscript and approved the final version of the manuscript. All authors contributed the clinical care for the patient in specialized settings.

Declaration of competing interest

None of the authors has any conflicts of interest or any financial ties to disclose.

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