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Case Report

Endobronchial hamartoma resected via bronchoscopy using high-frequency electrosurgical snare–Preoperative strategies using virtual bronchoscopy ☆,☆☆

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ABSTRACT

Pulmonary hamartomas are common benign lung tumors; however, endobronchial hamartomas are relatively rare. We report a case of asymptomatic endobronchial hamartoma in a 51-year-old man. Chest computed tomography revealed a 10-mm protrusion in the right main bronchus. Preoperative virtual bronchoscopy (VBS) was performed; subsequently, minimally invasive bronchoscopic resection was safely performed under local anesthesia. The use of VBS is a useful treatment strategy and follow-up modality.

Introduction

Pulmonary hamartomas are common benign lung tumors mostly located peripherally, with an incidence of 0.025%-0.32% according to several necropsy studies [1]. Endobronchial hamartomas are rare, accounting for only 1.4% of pulmonary hamartomas [2]. Peripheral hamartomas are often diagnosed after surgical resection; obtaining a tissue specimen for pathological diagnosis is challenging because the surface of the tumor is composed of normal bronchial tissue and most of the tumor components are cartilaginous, which cause the biopsy

Abbreviations: CT, computed tomography; VBS, virtual bronchoscopy.

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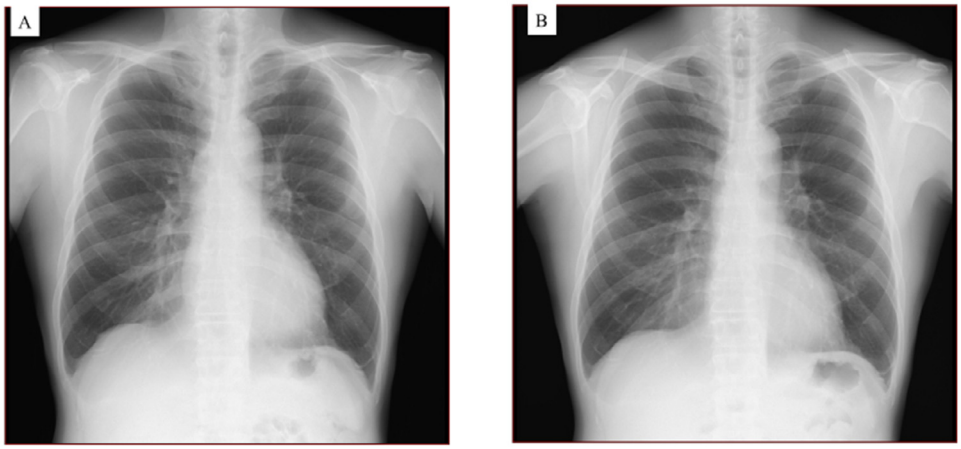

Fig. 1 – Chest radiography. (A) At admission. (B) After 1 year. There is no significant abnormality.

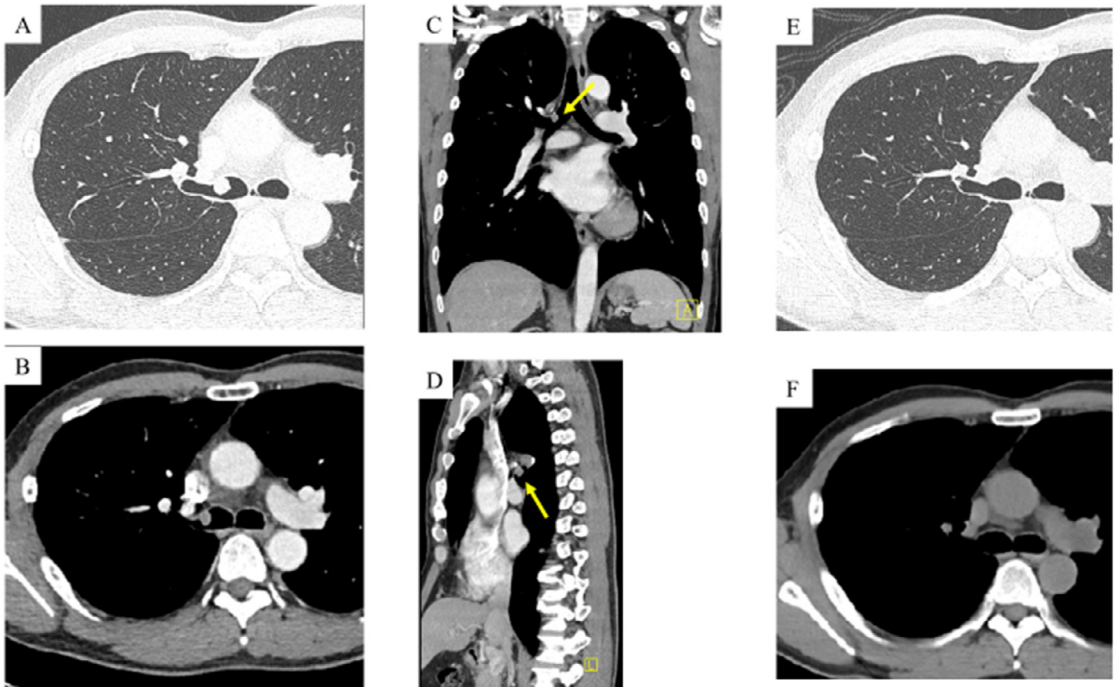

Fig. 2 – Chest computed tomography. (A–D) The axial, coronal, and sagittal views, show a pedunculated, elevated mass located ventral to the lateral side of the right main bronchus (yellow arrow). (E, F) One year after tumor resection, normal findings are observed.

forceps to slip [1]. However, since it is a benign disease, minimally invasive treatment is preferred.

We report a case of an endobronchial hamartoma in the right main bronchus that was safely resected via a single-stage bronchoscopy using a high-frequency electrosurgical snare under local anesthesia.

Case report

A 51-year-old Japanese man was referred to our hospital for specialist treatment of an endobronchial tumor. The patient had presented with cough and fever at the previous hospital. He had been diagnosed with right middle lobe pneumonia and treated with antibiotics. Chest computed tomography (CT) at presentation had revealed a polyp-like lesion in the right main bronchus. One month after the pneumonia was completely cured, a follow-up chest CT revealed similar findings; hence, the patient was referred to our hospital.

The patient had a history of a gastric submucosal tumor that was diagnosed and endoscopically resected at the age of 50.

The patient was a former smoker (5 pack-years); there was no significant family or allergy history. On admission, the patient had no remarkable respiratory symptoms.

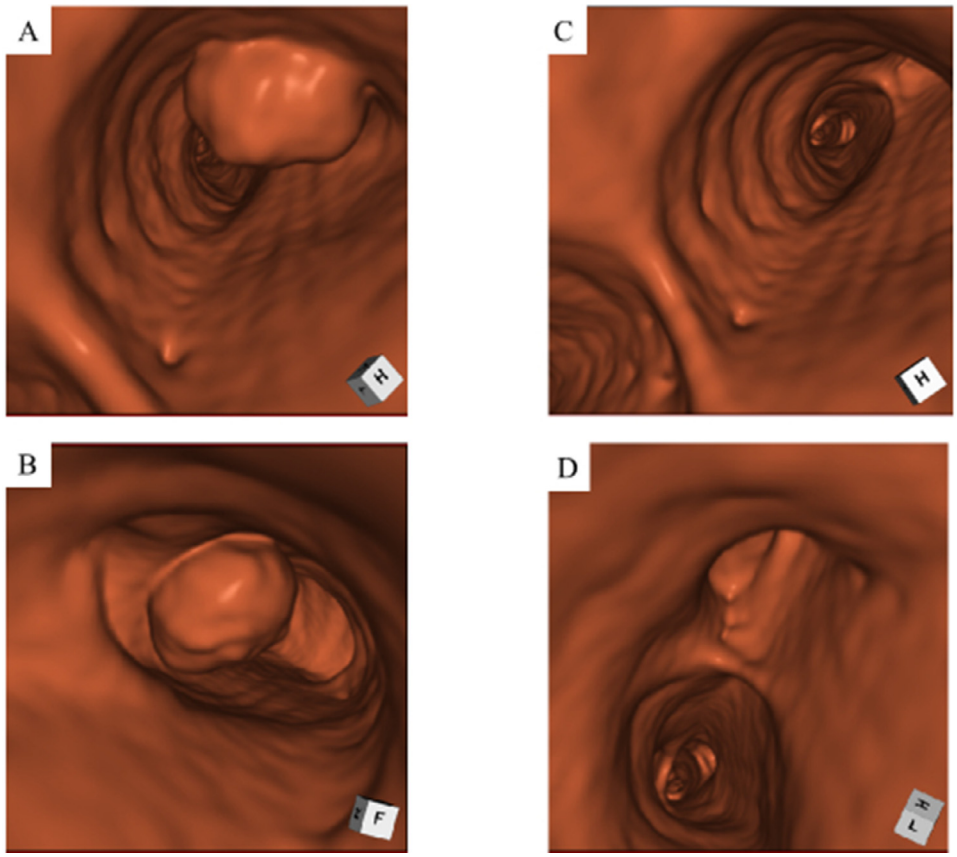

Fig. 3 – Virtual bronchoscopy. (A, B) A virtual bronchoscopy (VB) image is created from the thin slice image composition of high-resolution computed tomography; this confirms the presence of a pedunculated, smooth-surfaced lesion. VB shows a thin root of the tumor at the right bronchus from the distal side; hence, a treatment with endoscopic bronchoscopy is feasible. (A: antegrade approach, B: retrograde approach). (C, D) One year after tumor resection, a virtual bronchoscopy (VB) image was created from the thin slice image composition of high-resolution computed tomography images; the abnormality in the right main bronchus appears to have recovered (C: proximal view, D: distal view).

On clinical examination, his height and weight were 171 cm and 68 kg, respectively. He had a temperature of 36.8°C, blood pressure of 144/90 mmHg, pulse rate of 75 beats/min (normal sinus rhythm), respiration rate of 15 breaths/min, and oxygen saturation measured by pulse oximetry was 98% (room air). Chest auscultation revealed no abnormal breath sounds such as wheezing.

Laboratory tests revealed no abnormalities. Tumor markers were within normal range;

CYFRA 21-1 and carcinoembryonic antigen levels were 1.1 ng/mL and 2 ng/mL, respectively. Pulmonary function tests revealed FVC of 4.57 L, $FEV_{1.0}$ of 3.34 L, and $FEV_{1.0}$ /FVC ratio of 73.1%, and no obvious abnormalities.

Chest X-ray revealed no abnormal findings (Fig. 1A). Contrast-enhanced chest CT revealed a 10-mm protrusion in the right main bronchus. There was no calcification. The lesion demonstrated a slight internal contrast effect; however, its boundary adjoining the bronchial wall was clear (Figs. 2A-D).

Virtual bronchography was constructed from high-resolution thin slice CT images using Ziostation2 (Ziosoft, Inc, Tokyo, Japan). The protrusion was confirmed as a pedunculated lesion with a smooth surface. Based on this finding, a single-stage resection of the lesion using a high-frequency electrosurgical snare (Figs. 3A and B, Supplementary materials) was considered a feasible treatment option.

Bronchoscopy revealed a firm pedunculated tumor with a smooth surface located approximately 2 cm distal to the tracheal bifurcation (Figs. 4A and B). Endoscopic tumor resection was attempted using a flexible bronchoscope (BF type 1T260, Olympus), electrosurgical unit (ICC 350, ERBE), and high-frequency electrosurgical snare under local(2% lidocaine)and intravenous (midazolam) anesthesia. After endotracheal intubation (8.0 mm tube), the snare loop was used to pinch the stalk, and the tumor was resected at the root of the stalk with 2 cycles of energization at 30 watts for approximately 1-2 seconds each (Figs. 4C and D). There were no complications such as bleeding or respiratory failure. After tumor resection, the transected edge was extensively evaluated using brush cytology for the presence of tumor tissue; however, no abnormalities were found. The total treatment time was 30 m.

The resected tumor measured 10 × 7 × 5 mm. Histological examination revealed a layer of hyperplastic squamous epithelium, followed by hyaline cartilage, fatty tissue, and secretory glands (Fig. 5). The individual constituent cells

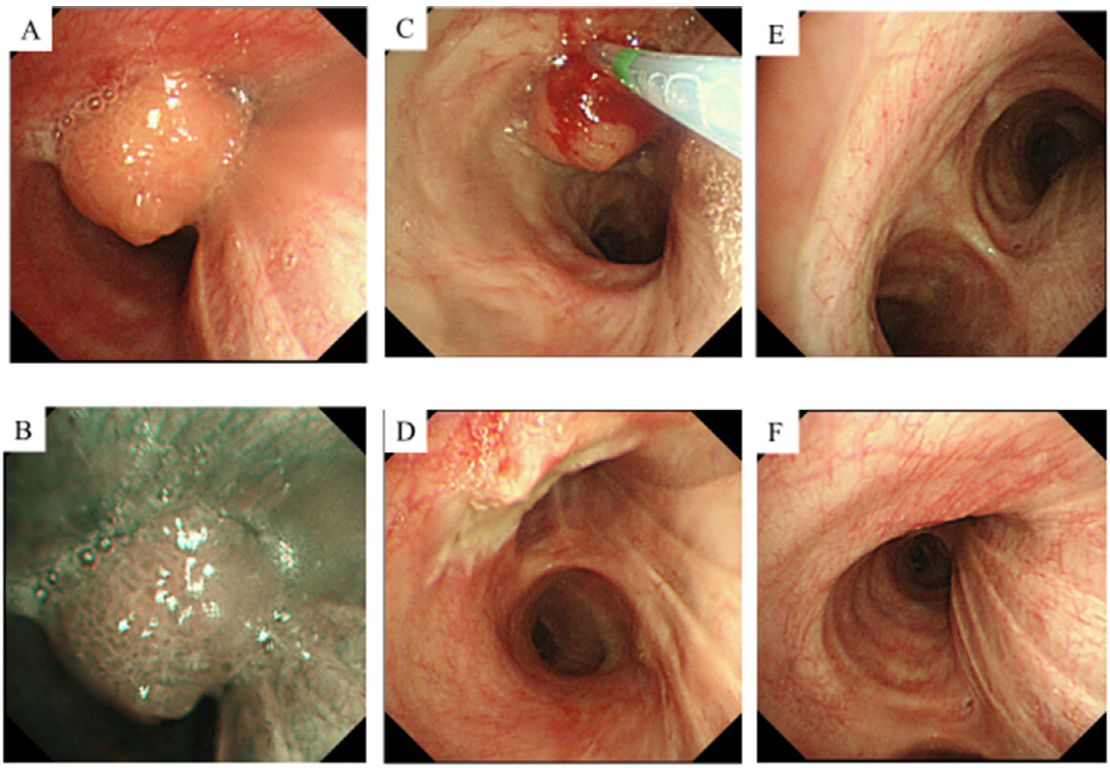

Fig. 4 – Flexible bronchoscopy findings. Bronchoscopy reveals a pedunculated polypoid lesion distal to the bifurcation of the right main bronchus. Its surface is smooth and no abnormal blood vessel development is observed. The lesion is safely resected using a high-frequency electrosurgical snare. (A) The pedunculated, smooth-surfaced tumor is located in the right main bronchus. (B) The surface structures and vessels observed in the tumor by narrow band imaging are similar to normal findings. (C) The tumor is completely resected using a high-frequency electrosurgical snare. (D) A view of the bronchial lumen after tumor resection. (E, F) Postoperative bronchoscopy shows a slight bronchial mucosal scar at the resection site.

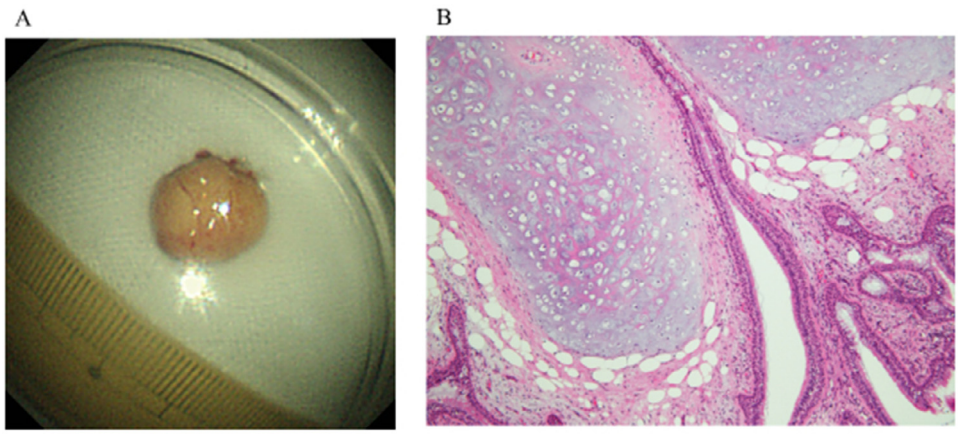

Fig. 5 – Pathological examination. (A) Macroscopic findings: smooth surface, normal epithelium, and glossy ball-like tumor. (B) Microscopic findings: hyperplastic squamous epithelium with no dysplastic cells and composed of fatty and bronchial gland tissues and hyaline cartilage.

were not atypical; hence, the tumor was diagnosed as an endobronchial cartilaginous hamartoma. The brush cytology result was negative; hence, it was considered a complete resection.

The postoperative course was uneventful, and the patient was discharged the following day. Bronchoscopy performed 3 months after discharge revealed only a slight scar on the mucosa at the resection site (Figs. 4E and F). After a follow-up pe-

Table 1. – Reported cases of endobronchial hamartoma resected with flexible bronchoscopy.

Author/year	Age/sex	Symptoms	Location (bronchus)	Bronchoscopy	HPE	Treatment	Relapse
Guarino et al. [8]	35/F	Cough, fever, Hemoptysis, SOB, chest pain	LUL	Smooth, reddish, poorly vascularized surface	Well-circumscribed and non-encapsulated cartilage nodules, respiratory epithelium, and fissure of fibrous tissue	Fenestrated crocodile biopsy forceps	No
Minalyan et al. [9]	49/M	Hemoptysis, SOB	LUL	White mass	Lipomatous hamartoma	Forceps debulking with cauterization.	No
Ahmed et al. [5]	53/M	Cough	LUL	Smooth mass	Hypercellular submucosal cartilaginous nodule with islands of cartilage.	Laser resection of the tumor and balloon dilation. LUL bronchus obstruction after 2 years and required LUL resection.	Yes
Mertoğlu et al. [10]	45/M	Cough, phlegm, fever	RMB & RBI	Mobile mass	Hamartoma	Endobronchial argon plasma coagulation and electrocauterization.	No
Sim et al. [11]	49/M	–	RLL	Pedunculated mass	Chondroid hamartoma	Cryotherapy with cryodebridement via flexible bronchoscopy.	No
	53/M	–	LLL (LB6)	Pedunculated mass	Chondroid hamartoma	Cryotherapy with cryodebridement via flexible bronchoscopy.	No
Gayathri Devi [12]	65/M	Cough, SOB	RUL	Polypoidal tumor	Fibrocollagenous tissue and mature fat cells.	Conservative. Asymptomatic 2 years post-bronchoscopy.	No
Poonja et al. [13]	56/F	Incidental	LLL	LLL bronchus tumor	Hamartoma	Polypectomy snare with electrocautery excision.	No
Yildirim et al. [14]	57/M	SOB, chest pain	LMB	Pedunculated mass	Gray, lobulated, elastic cartilaginous mucosal tissue.	Electrocautery by flexible bronchoscopy.	No
Mondello et al. [15]	65/M	Incidental	LMB	Pedunculated mass	Hamartoma	Flexible bronchoscopy and electrosurgical snaring.	No
Rai et al. [16]	40/M	Cough, fever, anorexia	LMB	Pink pedunculated mass	Disorganized lobules of cartilage and adipose tissue.	Resected endoscopically by diode laser.	No

F, female; M, male; HPE, histopathological examination; LLL, left lower lobe; LMB, left main bronchus; LUL, left upper lobe; RIB, right intermedius bronchus; RLL, right lower lobe; RMB, right main bronchus; RUL, right upper lobe; SOB, shortness of breath.

riod of 1 year, the patient has remained disease-free (Fig. 1B, Figs. 2E and F, 3C and D).

Discussion

"Hamartoma" is a term first used by Albrecht, in 1904, to describe tumor-like malformations resulting from probable developmental abnormalities. Subsequently, in 1934, Goldsworthy applied this term to benign lung tumors that predominantly comprised a combination of fatty and cartilaginous tissues [3].

Therefore, pulmonary hamartomas are benign malformations of lung tissues, with necropsy studies reporting an incidence of 0.025%-0.32% [3,4]. Most hamartomas occur in the periphery of the lungs. The tumors appear as solitary pulmonary nodules on radiography with an uneven density. Conversely, endobronchial hamartomas are rare benign neoplasms of the tracheobronchial tree that may present as endobronchial lesions, are seldom reported, and their characteristics are not well-elucidated. In a large review study (n = 215), only 1.4% of hamartomas were reported to be endobronchial [2].

In another study, most patients were asymptomatic; however, endobronchial hamartomas frequently produce respiratory complaints and radiographic abnormalities due to their location [3]. A literature review supports including endobronchial hamartomas in the differential diagnoses of patients with symptoms of wheezing, stridor, cough, dyspnea, atelectasis, hemoptysis, or recurrent pulmonary infections [2].

Histologically, a hamartoma is a mixture of mature mesenchymal tissue, such as adipose tissue, cartilage, bone, or smooth muscle bundles, and fibromyxoid tissue, with varying proportions of each component [5]. The surface of the tumor is composed of normal bronchial tissue, which causes biopsy forceps to slip, and the cartilaginous nature of the tumor component complicates biopsy tissue acquisition. Therefore, a multidisciplinary diagnosis using CT, MRI, and bronchoscopy is necessary.

CT is the imaging modality of choice to identify the pathognomonic signs of intralesional fat and calcifications. Bronchial hamartomas typically contain fat (HU of -40 to -120), calcifica-

tion (HU of >200), or a combination of both [6]. The reported prevalence of calcification and fat in hamartomas varies from 5% to 50% and 60% on CT, respectively [6]. The fat components may be localized or generalized within the nodule. Therefore, thin slice CT is essential to avoid missing the small foci of fat.

Owing to technological advances in bronchoscopy performance and treatment techniques, the primary treatment approach for endobronchial hamartomas is airway evaluation via flexible bronchoscopy with further interventions, including polypectomy with laser, electrocautery, or argon plasma coagulation [1]. Extensive procedures such as surgical wedge resection, lobectomy, or pneumonectomy may be performed depending on the location of the hamartoma and the extent of postobstruction damage. Treatment options from the 1900s to 2010 included invasive surgery such as bronchotomy, segmentectomy, lobectomy, and pneumonectomy [7]. However, the treatment of endobronchial hamartoma has advanced; currently, it is less invasive with the use of bronchoscopic excision with argon plasma coagulation, electrocautery, and tumor debulking with a flexible cryoprobe (Table 1) [5,8–16].

Virtual bronchoscopy (VBS) is a novel CT-based imaging technique that allows a noninvasive intraluminal evaluation of the tracheobronchial tree. Several studies have reported that VBS can accurately display the tracheal lumen and diameter [17]. The morphology of the carina can be accurately evaluated and the images are similar to those seen on fiberoptic bronchoscopy. In the present case, 3-dimensional (3D) image reconstruction using VBS enabled confirmation of the shape of the lesion and accorded a multidirectional overall view of the image (top, bottom, left, right, and 360º) [18].

The lesion was considered a benign tumor because of its smooth surface and peduncle; its shape was considered feasible for single-stage bronchoscopic resection. The contrast effect of the lesion was poor and the risk of bleeding was assumed to be relatively low. Thus, the tumor was completely resected via bronchoscopy in a single attempt using only a high-frequency electrosurgical snare, without the need for additional argon plasma coagulation.

The prognosis of endobronchial hamartoma is considered favorable; however, the recurrence rate after bronchoscopic resection is reported to be approximately 10% [19]. There is no consensus regarding the follow-up interval and frequency; long-term follow-up has revealed no evidence of malignant transformation of hamartoma in most cases [20]. In the present case, there was no evidence of recurrence by bronchoscopy and chest CT at 3 months and 1 year postoperatively, respectively; however, we will continue to observe the possibility of recurrence, by CT.

In conclusion, we believe that VBS is a promising and useful technique for noninvasive examination in planning treatment strategies and in post-treatment follow-up, which may be used in daily clinical practice.

Patient consent

Written consent for publication of the case and images was obtained from the patient.

Author contributions

M.S. was responsible for image acquisition and writing and editing the manuscript.

M.S. and H.W. managed the patient. M.S. and H.W. were responsible for the interpretation of data and drafting the manuscript. M.H. and S.I. assisted during the bronchoscopy procedure.

G.N., M.I., S.I., and Y.T. participated in the discussion regarding the treatment. M.H. and H.S. reviewed and supervised the manuscript.

All authors contributed significantly to the manuscript and approved its final version.

Supplementary material

A virtual bronchoscopy (VB) movie showing the tumor observed from the proximal and distal viewpoints.

This facilitates the understanding of the four-dimensional tumor structure, enabling the formulation of a pre-treatment simulation.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2022.08.018.

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