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Successful Resection of locally infiltrative Glomus Tumor without pulmonary resection

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ABSTRACT

INTRODUCTION: Extracutaneous glomus tumors occurring in the bronchus is very rare. Complete resection is basic procedure for treatment of glomus tumor. We present a glomus tumor of the left main bronchus that was successfully treated with rigid bronchoscopy followed by sleeve resection of the left main bronchus.
PRESENTATION OF CASE: A 56-year-old man underwent two term resections to glomus tumor that originated from the left main bronchus. Firstly, we performed palliative resection with rigid bronchoscopy to make the correct diagnosis and evaluate the extent of the tumor. We subsequently performed curative resection. No complications or recurrence has occurred since the operation took place one year ago.
DISCUSSION: Before curative resection, it is important to confirm the diagnosis and spread of the tumor. Therefore, palliative tumor resection by rigid bronchoscopy was useful to make the correct diagnosis, evaluate the extent of the tumor and open the bronchial lumen. After bronchoscopic treatment, curative pulmonary resection was performed and preservation of lung function was successful.
CONCLUSION: Two term resections enabled us to make an accurate diagnosis and evaluation, thereby preserving respiratory function without pulmonary resection.

1. Introduction

Glomus tumors are uncommon benign neoplasms originating in modified smooth muscle cells of the glomus body [1]. Extracutaneous glomus tumors occurring in the bronchus is very rare. In general, the treatment for glomus tumor is to perform a complete resection. Here we present a glomus tumor of the left main bronchus that was successfully treated with rigid bronchoscopy followed by sleeve resection with primary reconstruction of the left main bronchus. This work has been reported in line with the SCARE criteria [2].

2. Presentation of case

A 56-year-old man was admitted to a regional hospital due to cough and dyspnea. He was diagnosed as having bronchial asthma seven days before this episode. He had no history of systemic illnesses, smoking, and did not have any other symptoms. Physical examination and routine laboratory tests showed normal results. A chest CT scan was performed, showing a 1.5 × 0.9 cm polypoid lesion in the membranous portion of the left main bronchus and irregular thickness in the bronchial wall, and the tumor was extended to outside of the bronchial wall (Fig. 1a,b). Bronchoscopic findings showed a polypoid tumor over the posterior wall of the bronchus obstructing approximately 90% of the lumen (Fig. 2a). The surface of the tumor was smooth and covered with normal bronchial mucosa. From the visuals obtained of the tumor, a bronchial carcinoid tumor was suspected. The patient was referred to our hospital for further diagnosis and surgical treatment.

Pulmonary function tests revealed a slightly obstructive ventilation pattern (FEV1.0 = 3160 ml) mostly due to obliteration of the left main bronchus. Therefore, we planned a two-stage surgery. Firstly, the tumor resection with rigid bronchoscopy was planned to obtain a correct diagnosis and further information about the distal trun-

Abbreviations: FEV, forced expiratory volume; PA, pulmonary artery.

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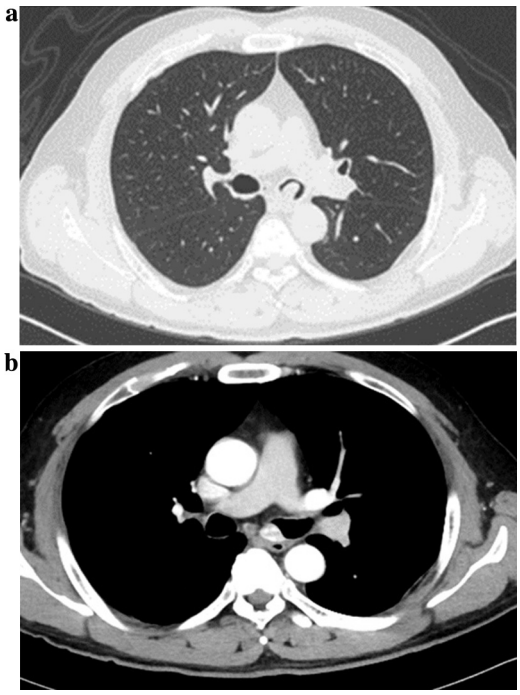

Fig. 1. Chest CT on admission.
Chest CT showed a polypoid tumor arising from the membranous portion of the left main bronchus (Fig. 1a,b).

cus intermedius. This was then followed by planning a curative resection. Under general anesthesia, partial resection of the tumor obstructing the distal part of the bronchus was performed with rigid bronchoscopy (Fig. 2b).

Two months after rigid bronchoscopy, we performed a curative resection. Although we adapted the approach of postero-lateral thoracotomy thorough the fifth intercostal space, we had concerns about maintaining the surgical view and operating space. We thus taped the left main pulmonary artery and aorta, then dissected the systemic lymph nodes in the perihilar, subaortic and subcarinal areas before performing the isolated sleeve resection. Following resection, we pulled the aorta and PA outside such that it did not obstruct our access. The airway was reconstructed by an end-to-end anastomosis with simple interrupted 3–0 monofilament absorbable sutures. We confirmed intra-operatively that the bronchial stump was tumor-free. The operation duration was 263 min, and the total volume of blood lost amounted to 100 ml. On pathological examination, the tumor was found to be a remnant glomus tumor (Fig. 3a,b). There were no lymph node metastases.

The patient had an uncomplicated postoperative course and was soon discharged in stable condition. He had no further respiratory complaints and the regular follow-up bronchoscopy revealed no signs of any anastomotic stenosis (Fig. 2c).

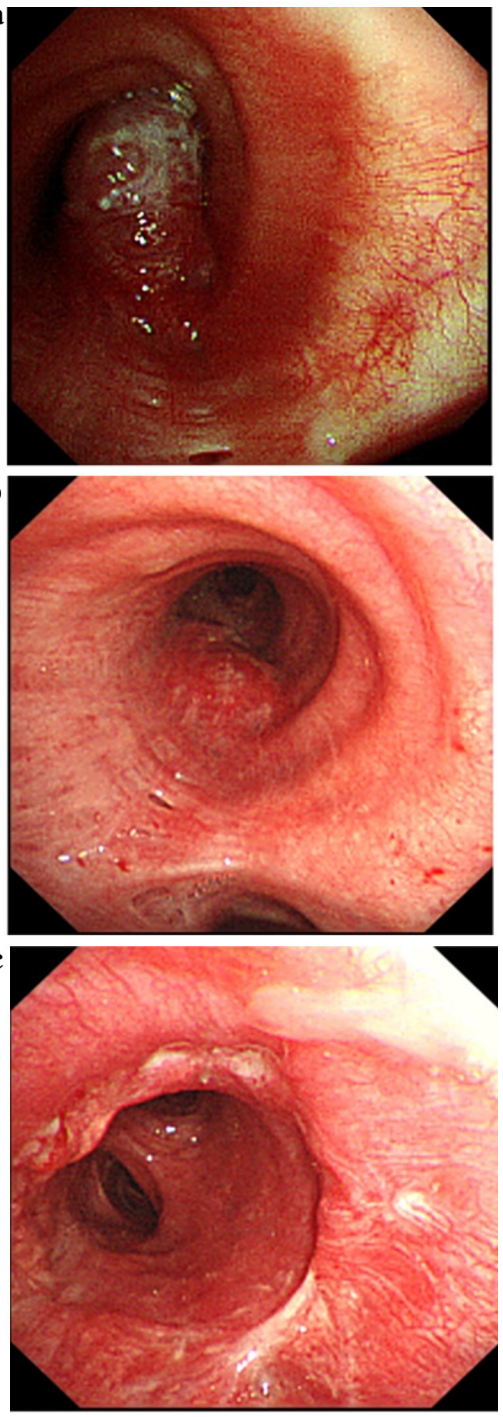

Fig. 2. Bronchoscopic findings.
Preoperative bronchoscopic image, the tumor almost completely obstructing the left main bronchus (Fig. 2a). After removal of the tumor by rigid bronchoscopy (Fig. 2b). Bronchoscopic image, one month after operation (Fig. 2c).

3. Discussion

Glomus tumor arising from the bronchus is a very rare occurrence. Only ten cases thus far, including our case study, have been published in the English-language literature [3–5]. Here we report a rare case of glomus tumor arising from the left main bronchus. We performed partial resection using rigid bronchoscopy to make a correct diagnosis and evaluate the extent of the tumor before performing sleeve resection.

In general, 95% of such tumors are benign and would therefore only require conservative treatment [5]. However, it is possible in rare cases for the tumor to be malignant. Among the various criteria, the primary indicators of tumor malignancy are “large size”, “deep location”, “atypical mitotic figure”, and “nuclear atypia”, but “infiltrative growth pattern” is considered unimportant [6]. In our case, there was a local infiltration point but no distant metastasis and no marked nuclear atypia were observed. So,“deep location” was cause for concern. Due to the possibility of malignancy, careful diagnosis of the tumor was warranted.

Endobronchial therapy is not preferred as the presence of any residual tumor increases the risk of recurrence without complete recognition. In our case, we had strong suspicions that the tumor

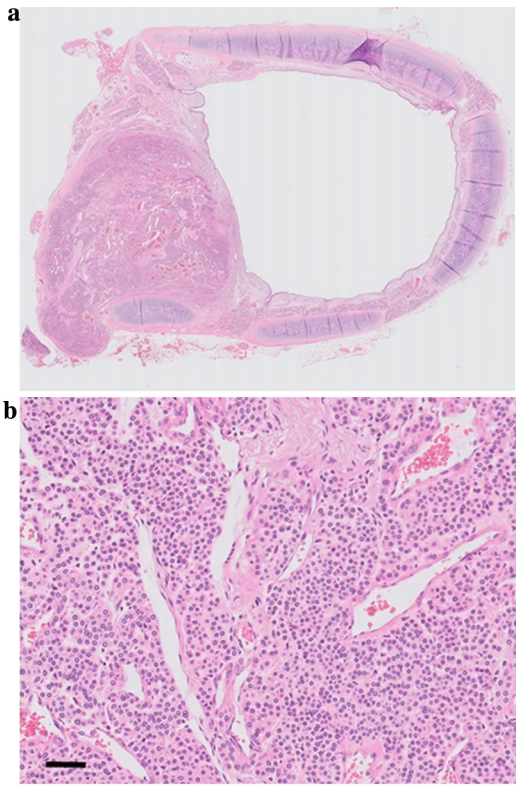

Fig. 3. Pathological findings.
Pathological examination. A well-circumscribed tumor in the bronchial wall (Fig. 3a), and tumor tissues were composed of dilated, cavernous hemangioma-like vessels, surrounded by clusters of small, uniform, and rounded cells with a round nucleus and amphophilic to lightly eosinophilic cytoplasm (Fig. 3b). Scale bar: 2 mm (Fig. 3a); 50 μm. (Fig. 3b)

had extended into the airway wall, leaving surgical resection as the only form of curative treatment. However, palliative resection with rigid bronchoscopy was first performed due to acute airway obstruction and the clinical diagnosis of the tumor with possible malignant potential. As such, we planned a two term resection. Resection with rigid bronchoscopy permitted us to make a correct diagnosis and evaluate the extent of the tumor before performing the sleeve resection. We previously reported that flexible bronchoscopy without general anesthesia was less invasive for patients. However, rigid bronchoscopy is preferred in the context of central or proximal airway tumors as it allows us to safely perform the procedure in the event of massive bleeding or hypoxia during the manipulation [7].

With regard to surgical operation, a bronchoplastic procedure should always be considered when the tumor arises from a main bronchus. In this case, the location and anatomy of disease was suitable for a bronchoplastic procedure. Therefore, our patient underwent a sleeve resection of the left main bronchus with an end-to-end anastomosis, checking the bronchial stumps to get clear margins. Before the resection, we confirmed the diagnosis and spread of the tumor, and were thus able to determine the excision range without the need for a wide free margin.

With these points in mind, we decided to take the approach of posterolateral thoracotomy as this tumor did not involve the carina and the structures in the subaortic space. The resection was complete with clear margins and lymph nodes. It was essential that we taped the aorta and performed dissection of lymph nodes before excision. This process ensured a good surgical view and sufficient space to carry out the operation. Follow-up with the patient since the operation one year ago has been unremarkable.

4. Conclusion

Although glomus tumors are mostly benign, correct diagnosis and identification of the location is important because of the malignant potential, and possibility of airway obstruction and bleeding, which may be life-threatening. For these reasons, palliative tumor resection by rigid bronchoscopy was useful to make the correct diagnosis, evaluate the extent of the tumor and open the bronchial lumen. After bronchoscopic treatment, curative pulmonary resection was performed and preservation of lung function was successful.

Conflict of interest

None.

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Ethical approval

Not applicable.

Consent

Written and signed consent from the patient to publish a case report has been obtained.

Author contribution

Tomohiro Obata, Takuro Miyazaki made study concept. Naoya Yamasaki, Tomoshi Tsuchiya, Keitaro Matsumoto, Go Hatachi, Yuka Kitamura collaborate in medical care. Kazuhiro Tabata examined pathology. Takeshi Nagayasu looked over the manuscript.

Guarantor

Tomohiro Obata.

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