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Therapeutic rigid bronchoscopy for endobronchial glomus tumors: a case series

Rong Lih Ho[1†], Byeong-Ho Jeong[2†], Joungho Han[3] and Hojoong Kim[2*]

Abstract

Background Glomus tumors (GTs) are rare, comprising only 2% of all soft tissue tumors. Pulmonary GTs are exceptionally rare, with fewer than 80 cases reported to date. Little is known about the therapeutic outcomes of rigid bronchoscopy for endobronchial GT.

Methods This is a case series of four patients with endobronchial GT who underwent therapeutic rigid bronchoscopy between February 2021 and June 2024.

Results The ages of the patients in our series ranged from 32 to 75 years, and all patients were male. Cough and blood-tinged sputum were present in all patients with endobronchial GT. The tumor sizes ranged from 1 to 3 cm. Complete endoscopic resection and laser cauterization via rigid bronchoscopy were achieved in two patients. One patient had incomplete resection of a 3-cm tumor in the segmental bronchus that showed radiological evidence of bronchial wall invasion. This patient subsequently underwent lobectomy seven months after bronchoscopic resection. The fourth patient was lost to follow-up. There was no mortality throughout the follow-up periods that ranged from 2.8 to 42.5 months. Factors favoring successful rigid bronchoscopy resection for endobronchial GT include a benign tumor in the central airways without bronchial wall invasion.

Conclusion Endoscopic resection and laser cauterization using rigid bronchoscopy may be a viable option for patients with endobronchial GT when surgery is not practical.

Clinical trial number Not applicable.

Keywords Endobronchial glomus tumor, Rigid bronchoscopy, Endoscopic resection, Laser cauterization

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Background

Glomus tumors (GTs) are rare, comprising only 2% of all soft tissue tumors [1, 2]. These tumors originate from the neuro-myo-arterial glomus body, which primarily functions in thermoregulation [1, 2]. The glomus body was first described by Masson in 1924, and Lumley and Stansfeld reported the first case of a clinically atypical infiltrating GT in 1972 [3–5]. Currently, GTs are classified into three categories: benign, malignant, and uncertain malignant potential [6].

Pulmonary GTs are exceptionally rare; only 40 tracheal and 36 bronchopulmonary GTs have been reported [1, 2]. Based on previous reports, open and endoscopic surgical resection are the mainstays for treatment of pulmonary GTs [1, 4, 7, 8]. Approximately 65% of patients with tracheal GTs who underwent surgical resection remained disease-free during the follow-up period, which ranged from 4 to 72 months. One patient succumbed to sepsis, while no documented outcomes were available for the remaining cases [4, 8]. One case was reported with airway stenosis at the anastomosis site following surgical resection, caused by granulation tissue formation and required further endoscopic removal [7].

Excision of endobronchial GTs using rigid bronchoscopy may be a viable option, especially for benign GTs [9]. Moreover, rigid bronchoscopy is a less invasive procedure and eliminates the risk of anastomosis site stenosis caused by granulation tissue, which can occur with surgical resection. However, available data on this treatment method are limited, and the majority of these data is case reports. To the best of our knowledge, this case series represents the first case series of endobronchial GTs treated with rigid bronchoscopy.

Methods

Patients

We retrospectively reviewed data from all patients who underwent rigid bronchoscopy for endobronchial GT at Samsung Medical Center from February, 2021 to June, 2024. This medical center is a 1979-bed tertiary care referral hospital in Seoul, South Korea, and is the largest and most active referral hospital for bronchoscopic interventions in the country [10]. The study was approved by the Institutional Review Board of Samsung Medical Center (IRB no. 2024-08-125). Informed consent was waived due to the retrospective nature of the study.

Bronchoscopic procedure

Rigid bronchoscopy was performed according to the standard techniques described by Jeong BH et al. [11]. In summary, patients were intubated with a rigid bronchoscope (Karl-Storz, Tuttlingen, Germany or Bryan Co., Woburn, MA, USA) after induction of general anesthesia. A flexible bronchoscope (BF 1T260 Olympus Corporation, Tokyo, Japan) and other adjunct tools were introduced through the rigid bronchoscope.

A GT typically presents as a round mass with a broad base. During treatment, the margin between the tumor and the normal mucosa was initially cauterized using a diode laser (Biolitec, Ceralas, Jena, Germany). The tumor was then mechanically debulked and resected using the rigid bronchoscope, followed by further cauterization of the tumor base to control the bleeding. The diode laser was operated at 14 watts, with a fraction of inspired oxygen maintained less than 40% during the laser cauterization. This process removed at least most of the tumor and maintained hemostasis.

Data collection and statistical analysis

We collected baseline data on patient demographics, tumor characteristics including computed tomography (CT) and bronchoscopy results, procedure details, and clinical outcomes. Using renal cell carcinomas as a reference [12], GTs showing 80 Hounsfield units (HUs) or more on contrast-enhanced CT were classified as high contrast enhancement. For data analysis, continuous variables are presented as range, and categorical variables are presented as number (percentage).

Results

Baseline characteristics

During the study period, four patients underwent rigid bronchoscopic endobronchial GT treatment. The demographic data, CT findings, and gross appearance of the tumors are summarized in Table 1. All four patients were transferred to our hospital after undergoing bronchoscopic biopsy at their previous hospitals. Of these, only one (case #4) was diagnosed with a glomus tumor, and the remaining three were transferred to our hospital without diagnostic results. At our hospital, rigid bronchoscopic procedures were performed immediately without additional bronchoscopic biopsy. The ages of the patients in our series ranged from 32 to 75 years, and all were male. Although two patients had a history of smoking, none had underlying lung disease. All four patients had normal ranges of platelets, prothrombin time, activated partial thromboplastin time, and normal liver and kidney functions. And none of the patients were taking antiplatelet drugs or anticoagulants before the procedure. All patients experienced blood-tinged sputum and cough, and dyspnea was observed in one case (case #1).

Among three patients who underwent contrast-enhanced thoracic CT before the procedure, two had contrast-enhanced tumors. The enhancement increase post-contrast for GTs ranged from 11 to 74 HU, and the tumor size ranged from 1 to 3 cm. In case #1, contrast-enhanced CT was not performed before the procedure. Case #2 showed signs of tumor invasion into

Table 1 Baseline characteristics of four patients with glomus tumor

No.	Year of visit	Age (years) /Gender	Comorbidity / Smoking Status	Symptoms	Location of lesion	Size (cm)/ Attenuation[a], HU	Gross form
#1	2021	46/M	Narcolepsy/ Active smoker, 25PY	C/D/B	Right main bronchus (proximal, posterior wall)	2.1/ 43 → NA	Round mass with a stalk
#2	2021	66/M	No/ Never	C/S/B	LB6 (bronchial invasion)	3.0/ 31 → 42	Round mass with broad base
#3	2021	75/M	No/ Never	C/S/B	Left main bronchus (proximal, medial wall)	2.1 / 28 → 95	Round mass with broad base
#4	2024	32/M	Hypertension/ Ex-smoker (12PY)	C/B	Left main bronchus (distal, lateral wall)	1.0/ 38 → 112	Lobulated mass with broad base

HU = Hounsfield unit, M = male, PY = pack-year, C = cough, D = dyspnea, B = blood-tinged sputum, NA = not available, S = sputum, LB6 = superior segmental bronchus of the left lower lobe

[a] These are the attenuation values of the tumors on computed tomography before and after contrast agent injection

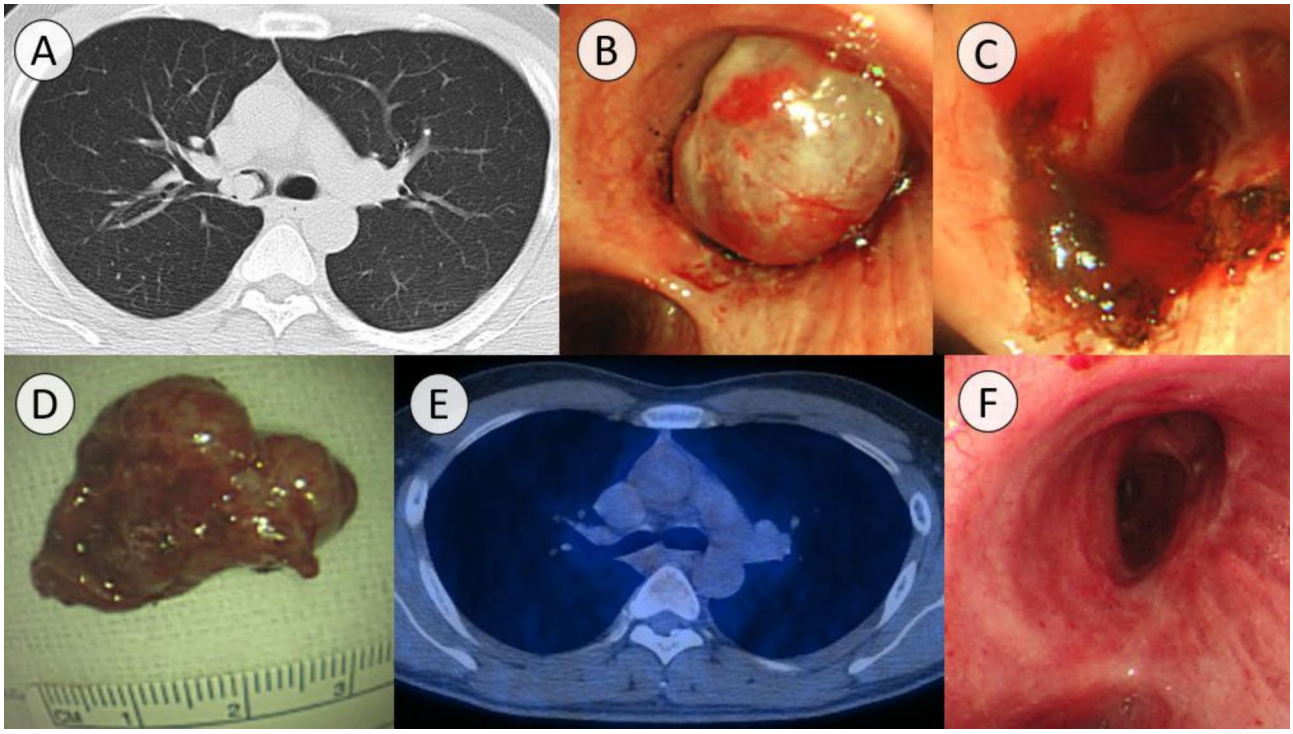

Fig. 1 Representative case (case #1). **(A)** A 46-year-old man underwent non-contrast computed tomography due to progressive dyspnea and hemoptysis. A round nodule was seen at the proximal right main bronchus. **(B)** Gross bronchoscopic findings were a round mass at the proximal right main bronchus causing near total occlusion. **(C)** After tumor resection using rigid bronchoscopy, the tumor base was cauterized using a diode laser. **(D)** The tumor mass was resected en bloc. Actin staining was diffusely strongly positive with Ki-67 negativity. **(E)** Positron emission tomography scan performed one week after the procedure showed complete tumor resection. **(F)** Surveillance bronchoscopy performed seven months after the procedure demonstrated no tumor recurrence. There was no recurrence during 42.5 months of follow-up

the bronchial wall, but case #3 and case #4 exhibited no such invasion. None of the cases demonstrated lymph node metastasis. Macroscopically, half of the tumors were round masses with broad bases protruding into the airway. However, that of case #1 was a round mass with a stalk, and that of case #4 was a lobulated mass with a broad base.

Treatment details and clinical outcomes

The representative case (case #1) is shown in detail in Fig. 1. CT and bronchoscopic images of the remaining cases are provided in Online Supplemental Figs. 1–3. Pathological images of all four patients are shown in Online Supplemental Fig. 4. Tumor resection using a laser and rigid bronchoscope was effectively performed in all cases but one, case #2 (Table 2). In this patient, the 3.0-cm tumor was in the left lower lobe bronchus involved the segmental bronchus (LB6), leading to incomplete tumor resection. This patient was confirmed to have residual tumor tissue and required a lobectomy seven months after the procedure. In another patient, case #3, initial tumor resection was successful, but

Table 2 Treatment details and results

No.	Bronchoscopic intervention	Anesthesia time / Procedure time, min[a]	Additional treatment	Baseline FEV_1 and FVC, L (% predicted value)	Post-procedural FEV_1 and FVC, L (% predicted value)	Follow-up duration (month) and final patient status
#1	Debulking, laser	31 / 23	No	2.41 (55) / 3.91 (73)	4.04 (93) / 4.96 (93)	42.5 / Under follow-up
#2	Debulking, laser	43 / 23	Surgical resection[b]	2.22 (88) / 2.75 (80)	NA	42.5 / Under follow-up
#3	Debulking, laser	42 / 16	No, suspect recurrence[c]	1.75 (71) / 2.29 (64)	2.82 (112) / 3.07 (85)	2.8 / Lost to follow-up
#4	Debulking, laser	30 / 15	No	3.47 (87) / 4.25 (86)	3.59 (89) / 4.30 (87)	5.6 / Under follow-up

FEV_1 = forced expiratory volume in one second, FVC = forced vital capacity, NA = not available

[a] Anesthesia time refers to the time from the start of general anesthesia to awakening from general anesthesia. Procedure time refers to the time from rigid bronchoscopic intubation to extubation

[b] He underwent left lower lobectomy via video-assisted thoracoscopy surgery (VATS) seven months after rigid bronchoscopy. There was residual tumor tissue with a clear surgical margin in the surgical specimen

[c] Suspect recurrence because of the presence of a small nodule at the proximal left main bronchus on surveillance computed tomography at three months post-rigid bronchoscopy. No confirmational biopsy was performed on this patient, however, because he was lost to follow-up

subsequent surveillance thoracic CT revealed a small nodule at the proximal left main bronchus that raised concerns of tumor recurrence or granulation tissue formation. Unfortunately, no confirmatory tissue biopsy was performed as the patient was lost to follow-up. Three patients, cases #1, #3, and #4, showed post-procedural FEV_1 improvement (gains of 1.63 L, 1.07 L and 0.12 L, respectively) on spirometry. As of November 2024, three patients were being followed; the other was lost to follow-up. There were no intra-operative or post-operative complications. Bleeding was well-controlled following the resection, as the tumor margin was carefully cauterized before the resection, and the tumor base was immediately cauterized afterward to prevent further bleeding. No mortality occurred during the follow-up periods that ranged from 1.0 to 29.7 months.

Discussion

In their literature reviews, Venegas et al. and Oide et al. reported that tracheal and bronchopulmonary GTs predominantly affect male patients (65% and 72%, respectively); with 80% and 64% of cases occurring in individuals aged 40 and older [1, 2]. Consistent with these findings, our case series demonstrated that endobronchial GTs predominantly occur in male adults 30 to 70 years old. Additionally, all cases in our series involved otherwise healthy adults without underlying lung disease.

Endobronchial GT is usually symptomatic, unlike GTs located in the pulmonary parenchyma, which are often asymptomatic. Previous case reports on GTs in the respiratory system, either lung or airway, indicated that most bronchial and tracheal GT cases presented with either cough or hemoptysis; however, most lung GTs were asymptomatic [1, 13]. Similarly, cough and blood-tinged sputum were the predominant symptoms in all four of our cases.

GTs have variable and non-specific features on CT but typically appear as round, nodular lesions. Therefore, the common differential diagnoses for endobronchial GT include carcinoid tumor and adenoid cystic carcinoma [13, 14]. Our series demonstrated that the endobronchial GTs generally resulted in high contrast enhancement on CT. Additionally, most tumors were in the central airway with no evidence of lymph node metastasis on CT imaging and bronchoscopy. According to previous literature, three cases were resected using endoscopic Nd-YAG laser. Two of these patients remained disease-free after 12 and 24 months of follow-up, while the third required external radiation and no documented outcome was available [4, 8]. Hence, tumor resection and laser cauterization via rigid bronchoscopy might be a viable option for achieving tumor clearance. Besides that, GTs are highly vascular [14, 15], so laser cauterization at the margin between the tumor and normal mucosa is a crucial step to reduce the risk of excessive bleeding during tumor resection via rigid bronchoscopy. Thus, the risk of bleeding is higher compare to surgery if cauterization is not performed properly. However, rigid bronchoscopy is generally more readily accepted, especially by younger patients. Additionally, it eliminates the risk of post-resection stenosis commonly associated with surgery.

In general, GTs are benign neoplasms. However, a few cases have been reported to be malignant glomangiosarcomas with the potential for metastasis to mediastinal lymph nodes and other organs, particularly the gastrointestinal tract [16–18]. Therefore, histological confirmation and exclusion of any potential malignant transformation, such as nuclear atypia and high mitotic activity [1, 14], are essential before considering tumor

resection via rigid bronchoscopy. Regular surveillance using CT and bronchoscopy post-resection is probably sufficient. Our case #1 had no residual tumor or recurrence on surveillance after rigid bronchoscopy resection, demonstrating that complete endoscopic resection of endobronchial GT is possible. Our case #2 showed that complete resection via rigid bronchoscopy is not always feasible when a large tumor occupies the peripheral airway with bronchial invasion. This is a new finding in our case series, as previous literature has only reported endobronchial GTs treated with endoscopic resection located either in the trachea or main bronchus [3, 4, 8]. Our case #4 remained tumor-free during a short follow-up period. In summary, factors favoring successful rigid bronchoscopy resection for endobronchial GTs include benign tumors with a central airway tumor location without bronchial wall invasion.

This study has a few limitations. First, generalizability of the findings is limited as this was a retrospective case series from a single tertiary center with a small number of cases. However, given the rare occurrence of these tumors, a prospective study with a larger sample size is challenging. Second, there were difficulties in conducting a more comprehensive assessment due to missing data such as pulmonary function test and contrast-enhanced CT results and patient-reported outcomes. More data are needed to consider whether positron emission tomography scan should be included in baseline investigations to screen for metastasis. Finally, more evidence is needed to identify the optimal duration of follow-up as there is a lack of data on recurrence risk.

Conclusion

In conclusion, endoscopic resection followed by laser cauterization using rigid bronchoscopy may be a viable option for endobronchial GTs, particularly when the tumor is benign and located in the central airways without bronchial wall invasion. We hope that this study will assist clinicians in selecting patients who will benefit most from rigid bronchoscopic intervention.

Abbreviations

CT Computed tomography
HU Hounsfield units
GT Glomus tumor

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s12890-024-03466-z.

Supplementary Material 1

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None.

Author contributions

RLH has been involved in data curation, formal analysis, interpretation of data, and drafting the work. BHJ has been involved in conceptualization, data curation, formal analysis, interpretation of data, and drafting the work. JH has been involved in methodology, resources, and reviewing the work. HK has been involved in conceptualization, investigation, methodology, resources, and reviewing the work. All authors read and approved the final manuscript.

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Data availability

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

This study was conducted in accordance with the Declaration of Helsinki and approved by the Institutional Review Board of Samsung Medical Center (IRB no. 2024-08-125). Informed consent was waived by the IRB.

Consent for publication

Informed consent was waived by the IRB because it is a retrospective study, some patients are not being followed up, and it does not contain personal information that can be used to infer individual patients. However, we want to emphasize that we strictly adhered to ethical principles and ensured patient confidentiality throughout the study.

Competing interests

The authors declare no competing interests.

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