Received: 21 December 2022 | Accepted: 15 January 2023

DOI: 10.1002/rcr2.1091

CASE REPORT

Respirology Case Reports OPEN ACCESS APSR WILEY

Two cases of large tracheobronchial schwannomas completely resected by rigid bronchoscopy with multiple instruments

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Funding information
Jeju National University

Associate Editor: Phan Nguyen

Abstract

Primary tracheobronchial schwannomas are extremely rare. Surgical treatment has been the first choice for these benign tumours due to the substantial residual rates and recurrences after bronchoscopic resection. In addition, there has been limited information on bronchoscopic removal of endobronchial schwannomas. We describe two cases of large tracheal and bronchial schwannomas that were completely and successfully resected by snare electrocautery, insulation-tipped knife, and argon plasma coagulation under rigid bronchoscopy. These cases highlight that rigid bronchoscopic treatment with these multiple instruments can be a good treatment option for endotracheal or endobronchial schwannomas.

KEYWORDS
endobronchial tumours, rigid bronchoscopy, schwannoma

INTRODUCTION

Schwannomas are benign slow-growing tumours that arise from peripheral, spinal, or cranial nerves, excluding the optic and olfactory nerves.[1] They may occur almost anywhere in the body, but primary tracheobronchial schwannomas are extremely rare.[2,3] Surgical treatment has been the first choice for benign endobronchial tumours, but in recent years, bronchoscopic tumour removal has been reported to be effective.[4,5] However, bronchoscopic resection can be incomplete and consequently may lead to local recurrence. Data from case reports and small retrospective studies in the literature were also not sufficient to develop conclusive information on bronchoscopic removal of tracheobronchial schwannomas, and more results of the interventional bronchoscopic treatment need to be gathered. Therefore, we present two cases of large tracheal and bronchial schwannomas that were completely and successfully resected by snare electocautery, insulation-tipped (IT) knife, and argon plasma coagulation (APC) with rigid bronchoscopy.

Changhwan Kim and Hae-Seong Nam contributed equally to this work.

CASE REPORT

Case 1

A 72-year-old man presented to the outpatient department with an abnormality on a chest radiograph. On contrast-enhanced chest CT scan, a 1.4 × 1.1 cm size, heterogeneously enhancing endobronchial mass was found in the proximal left upper lobar bronchus (Figure 1A). Fiberoptic bronchoscopy showed the presence of a polypoid mass in the orifice of the left upper lobar bronchus causing a near-complete bronchial obstruction (Figure 1B). Bronchoscopic forceps biopsies were tried, but failed due to the hard and slippery surface of the tumour. Rigid bronchoscopy was performed to remove the endobronchial tumour and make a confirmatory diagnosis. Due to the polypoid shape and wide

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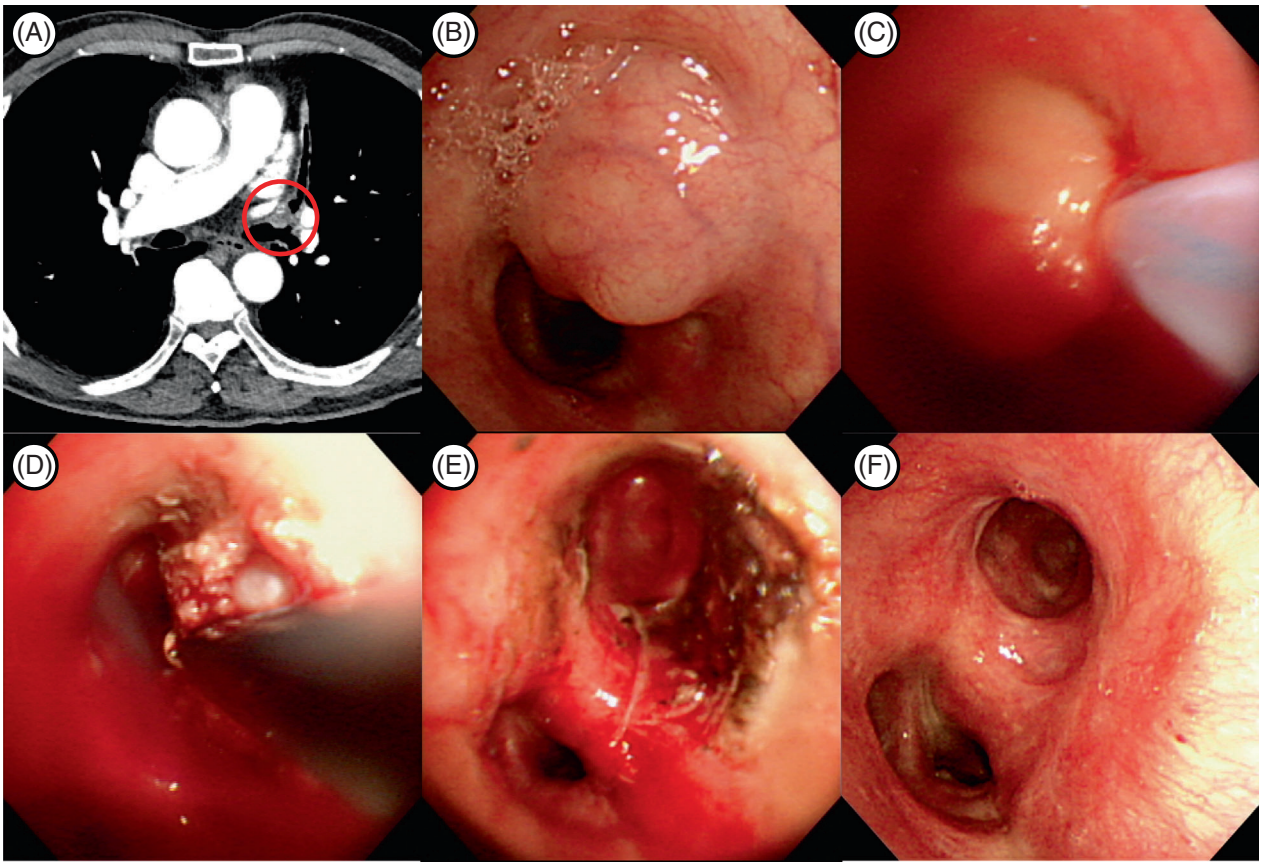

FIGURE 1 (A) A chest CT scan revealed a heterogeneously enhancing mass in the proximal left upper lobar bronchus. (B) Fiberoptic bronchoscopy showed the presence of a polypoid mass in the orifice of the left upper lobar bronchus causing a near complete obstruction. (C) The endobronchial mass was successfully removed by snare electrocautery, (D) insulation-tipped knife, (E) and argon plasma coagulation under rigid bronchoscopy. (F) Follow-up bronchoscopy revealed no recurrence at 5-year after initial treatment

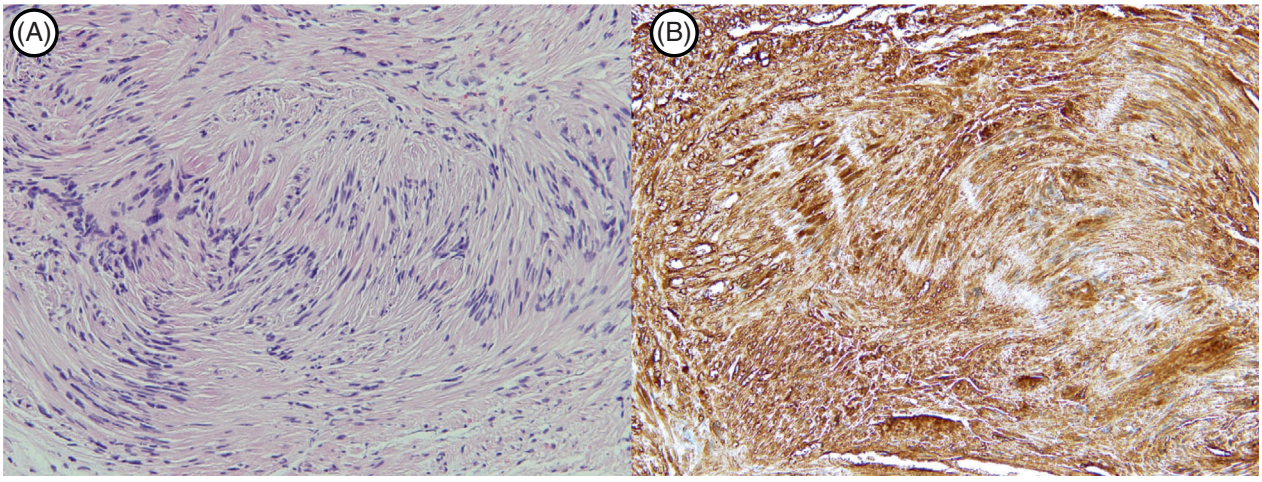

FIGURE 2 (A) The tumour was composed of interlacing bundles of spindle cells arranged in a palisading pattern without mitosis or necrosis (haematoxylin and eosin staining, ×200). (B) Immunohistochemistry revealed intense protein S-100 expression (×200)

base, we initially use snare electrocautery (ClearGrasp Snare, FINEMEDIX, Daegu, Republic of Korea) to cut the base of the tumour (Figure 1C). After that, the residual tumour at the base was resected with an IT knife (KD-610 L, Olympus, Tokyo, Japan) (Figure 1D). Finally, the possible remnant tumour cells below the base were coagulated and vaporized by APC (FiAPC Probe 2200 A, Erbe, Tübingen, Germany) (Figure 1E). The postoperative course was uneventful.

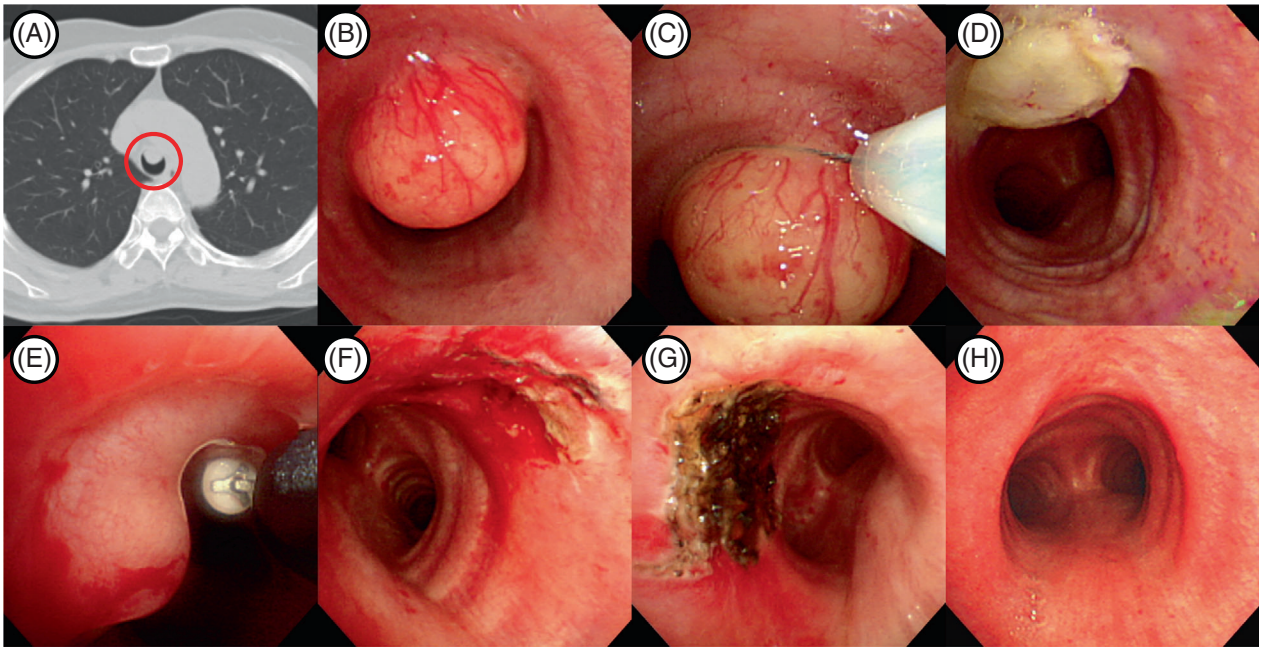

FIGURE 3 (A) A low-dose chest CT scan revealed a large ovoid endotracheal mass on the anterior wall of the mid-trachea. (B) Fiberoptic bronchoscopy showed the presence of a polypoid mass with a significant narrowing of the tracheal lumen. (C) The endobronchial mass was resected using snare electrocautery at the same time as flexible bronchoscopy, (D) but the base was not completely excised. (E, F) Rigid bronchoscopy was performed to remove the remnant tumour using an insulation-tipped knife, (G) and the resected tumour base was coagulated and vaporized by argon plasma coagulation. (H) Follow-up bronchoscopy revealed no recurrence at 2-year after initial treatment

Histologically, the tumour was confirmed to be schwannoma with positive S-100 protein by immunohistochemical staining (Figure 2). The patient was monitored in an outpatient clinic and there was no recurrence during the 5-year follow-up period (Figure 1F).

Case 2

A 60-year-old woman was referred to our hospital for an endotracheal tumorous lesion. A low-dose chest CT scan obtained from an outside hospital revealed a 1.6 × 1.1 cm, ovoid endotracheal mass on the anterior wall of the mid-trachea (Figure 3A). She complained of difficulty breathing, and airflow limitation was also detected by spirometer at the presentation. Fiberoptic bronchoscopy showed the presence of a polypoid mass with a narrowing of more than 50% of the tracheal lumen (Figure 3B). We attempted resection of the lesion using snare electrocautery to improve her dyspnea (Figure 3C), but the base was not completely excised (Figure 3D). Six weeks later, rigid bronchoscopy was performed to remove the remnant tumour using an IT knife (Figure 3E, F). Additionally, the resected tumour base was coagulated and vaporized by APC (Figure 3G). The postoperative course was uneventful. The tumour was also confirmed to be schwannoma with positive S-100 protein. The patient was monitored in an outpatient clinic and there was no recurrence for the 2-year follow-up period (Figure 3H). The airflow limitation was consequently resolved and the forced expiratory volume in 1 s increased from 1.94 L (78% predicted) to 2.23 L (90% predicted).

DISCUSSION

Primary tracheobronchial schwannoma is a rare disease. Bronchoscopic intervention or surgical resection can be used to treat primary tracheobronchial tumours. However, in general, surgical treatment is considered to be the first treatment for primary tracheobronchial tumours. Due to scarcity, the standard of care for tracheobronchial schwannomas has not been adequately established.[6]

Rusch et al. suggested that the choice of treatment may be influenced by the clinical presentation of the tumour (pedunculated or sessile), the risk of tracheal resection, and the presence or absence of an extratracheal component.[7] Hamouri et al. also suggested that, for tumours with a larger base (sessile type) or extraluminal lesion, and in those patients without surgical contraindications, surgical resection is a preferred option.[8]

In this case study, the shape of both the tracheal and bronchial tumours looked like domes with wide bases, which were approximate sessile rather than pedunculated. Although relatively in advanced age, these two patients were in good health and were not contraindicated to surgical treatment. Extraluminal extension was not clearly distinguishable in CT images of both cases. Surgical resection may be suitable for these cases according to

the reports mentioned above. However, complications following tracheal resection and end-to-end anastomosis are rare but can be devastating.[9] Complications such as atelectasis, bronchopleural fistula, or bronchial stenosis can develop after bronchial sleeve resection, and operative mortality is in the range of 1%–7%.[10] Meanwhile, in a recent multicentric retrospective analysis, the complication rate and mortality of rigid tracheobronchoscopy were very low in cases of non-malignant airway disease, performed as a scheduled elective procedure.[11] Therefore, surgical resection of tracheal or bronchial schwannomas should be decided with careful consideration and a comprehensive discussion about possible complications.

In the published literature on bronchoscopic intervention, various instruments and techniques have been used for schwannoma excision, such as electronic snaring, laser, APC, cryotherapy, and/or microdebrider under flexible or rigid bronchoscopy.[12] Approximately one fifth of cases with relapse were reported and were further treated by surgery for complete resection.[4,12] Although risk factors have not been studied yet, tumour recurrence is probably attributable to incomplete resection or extraluminal extension. If an extraluminal lesion is apparent on CT scan or magnetic resonance images, it would be highly undesirable to perform bronchoscopic resection. If an extraluminal lesion is not clearly visible, a bronchoscopic intervention can be selected for a radical cure.

To avoid incomplete resection, it is obviously better to perform rigid bronchoscopy under general anaesthesia with muscle paralysis, which guarantees immobilization and complete cough control, than flexible bronchoscopy under sedation. In the case of a polypoid tumour, a sizable volume of the tumour can be removed using snare electrocautery. However, unless pedunculated, cutting close to the base would not be easy with a simple snaring. The IT knife is an endoscopic instrument with a small ceramic ball attached as an insulator at the end of a needle knife, which is used mainly for endoscopic mucosal resection or submucosal dissection on the gastrointestinal track.[13] Although there have been only a few reports, they suggested that the use of an IT knife in bronchoscopic treatment was effective and could have a lower risk of distal airway injury due to the insulating tip and its lateral cutting capability.[14] Therefore, the remnant lesions after initial snare cutting can be effectively removed with an IT knife.

In the case of a benign tumour, the macroscopically invisible tumour tissues at the resected base are usually devitalized by laser or APC. APC is a noncontact thermal treatment using ionized argon gas jet flow to conduct electrons and has the strong advantages of being cheaper and easier to use compared to a laser without concern of biohazards.[15] Although the effects of APC are superficial compared with laser, deep tissue coagulation may not always be preferable, and APC after electrocautery is elegant even for the treatment of superficial squamous cell carcinoma of the early stage.[15] Therefore, the use of APC may also be beneficial for necrosis of the resected base of benign airway tumours.

Hence, we present two cases of large tracheal and bronchial schwannomas that were completely and successfully resected by rigid bronchoscopy with multiple instruments. In summary, local resection with multiple instruments such as snare electrocautery, IT knife, and APC under rigid bronchoscopy may be a preferable treatment option for patients with endotracheal and endobronchial schwannomas without definite evidence of extraluminal extension.

AUTHOR CONTRIBUTIONS

Changhwan Kim: Conception and design, acquisition of data, and drafting of the manuscript. **Hae-Seong Nam**: Supervision and conceptualization. **Yousang Ko**: Conceptualization and writing-review and editing.

ACKNOWLEDGMENT

We thank the allied health professions, especially the chief nurse Seunghee Kim, for their valuable work.

FUNDING INFORMATION

This work was supported by the 2022 education, research, and student guidance grant funded by Jeju National University.

CONFLICT OF INTEREST

None declared.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images. This study was approved by the Institutional Review Boards of the Jeju National University Hospital (No. 2022–11-020).

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REFERENCES

1. Zachariades N, Skoura C, Papageorgiou G, Chrissomali E. Giant ancient neurilemmoma of the cervical region: report of case. J Oral Maxillofac Surg. 2001;59(6):668–72.
2. Kasahara K, Fukuoka K, Konishi M, Hamada K, Maeda K, Mikasa K, et al. Two cases of endobronchial neurilemmoma and review of the literature in Japan. Intern Med. 2003;42(12):1215–8.
3. Suzuki H, Sekine Y, Motohashi S, Chiyo M, Suzuki M, Haga Y, et al. Endobronchial neurogenic tumors treated by transbronchial electrical snaring and Nd-YAG laser abrasion: report of three cases. Surg Today. 2005;35(3):243–6.
4. Jung YY, Hong ME, Han J, Kim TS, Kim J, Shim YM, et al. Bronchial schwannomas: clinicopathologic analysis of 7 cases. Korean J Pathol. 2013;47(4):326–31.
5. Jin B, Wang T, Wang J, Qiu X, Pei Y, Wang Y, et al. Interventional bronchoscopic therapy in adult patients with tracheobronchial schwannoma. Ann Palliat Med. 2021;10(6):6279–86.

6. Chen H, Zhang K, Bai M, Li H, Zhang J, Gu L, et al. Recurrent transmural tracheal schwannoma resected by video-assisted thoracoscopic window resection: a case report. Medicine (Baltimore). 2019;98(51):e18180.
7. Rusch VW, Schmidt RA. Tracheal schwannoma: management by endoscopic laser resection. Thorax. 1994;49(1):85–6.
8. Hamouri S, Novotny NM. Primary tracheal schwannoma a review of a rare entity: current understanding of management and followup. J Cardiothorac Surg. 2017;12(1):105.
9. Auchincloss HG, Wright CD. Complications after tracheal resection and reconstruction: prevention and treatment. J Thorac Dis. 2016;8(Suppl 2):S160–7.
10. Beauchamp G. Fundamentals of standard sleeve resection. Thorac Surg Clin. 2018;28(3):285–9.
11. Fortin M, Yarmus L, Rendina EA, Rafeq S, Andrade R, Michaud G, et al. Multi-institutional retrospective analysis of adverse events following rigid tracheobronchoscopy. Respirology. 2021;26(1):87–91.
12. Ge X, Han F, Guan W, Sun J, Guo X. Optimal treatment for primary benign intratracheal schwannoma: a case report and review of the literature. Oncol Lett. 2015;10(4):2273–6.
13. Ono H, Hasuike N, Inui T, Takizawa K, Ikehara H, Yamaguchi Y, et al. Usefulness of a novel electrosurgical knife, the insulation-tipped diathermic knife-2, for endoscopic submucosal dissection of early gastric cancer. Gastric Cancer. 2008;11(1):47–52.
14. Park JW, Ko Y, Kim C. Use of an insulation-tipped knife during rigid bronchoscopic treatment of benign tracheobronchial stenosis. Medicina (Kaunas). 2021;57(3):251.
15. Bolliger CT, Sutedja TG, Strausz J, Freitag L. Therapeutic bronchoscopy with immediate effect: laser, electrocautery, argon plasma coagulation and stents. Eur Respir J. 2006;27(6):1258–71.

How to cite this article: Kim C, Nam H-S, Ko Y. Two cases of large tracheobronchial schwannomas completely resected by rigid bronchoscopy with multiple instruments. Respirology Case Reports. 2023;11:e01091. https://doi.org/10.1002/rcr2.1091