CASE REPORT

A Case of Benign Tracheal Schwannoma Treated with Tracheal Resection

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ABSTRACT — Background. We here report a case of surgical tracheal resection for a rare benign tracheal schwannoma, also known as neurilemroma, and discuss the different therapeutic options. Case. A 47-year-old man presented to our hospital with progressive shortness of breath and bloody sputum. Bronchoscopy demonstrated a wide-based submucosal tumor on the membranous portion of the lower trachea. A tissue biopsy was performed and the tumor was diagnosed as benign schwannoma. Segmental resection of the trachea with a primary end-to-end anastomosis was successfully performed via right posterolateral thoracotomy. The postoperative course was uneventful, and at the latest follow-up, the patient was doing well and was recurrence-free. Conclusion. We encountered a case of tracheal schwannoma successfully treated with partial tracheal resection and end-to-end anastomosis. The appropriate treatment (surgery or bronchoscopic resection) for benign tracheal tumors should be carefully decided on the basis of the clinical findings.

(KEYWORDS — Tracheal schwannoma, Benign tracheal tumor, Tracheal resection)

INTRODUCTION

Tracheal tumors are rare, and have only infrequently been reported. Among the reported tracheal tumors, most were malignant, such as squamous cell carcinoma or adenoid cystic carcinoma, whereas benign tumors represent only a minority of these cases. As a therapeutic option, tracheal resection is considered rather invasive, and recently, the usefulness of bronchoscopic resection for benign tracheal tumors has been demonstrated. We here report a rare case of surgical resection for a benign tracheal schwannoma, and discuss the therapeutic options for this condition.

CASE REPORT

A 47-year-old man presented to our hospital with progressive shortness of breath and bloody sputum. He was a current smoker (50 pack-years) but had no history of pulmonary disease. His physical examination results were normal, and respiratory wheezing could not be heard. The chest roentgenogram findings appeared normal. The flow-volume curve showed slight abnormality but with an incomplete plateau line (Figure 1A). We did not confirm upper airway obstruction, but suspected the presence of some airway lesion from the results of this examination.

Computed tomography (CT) revealed an endotracheal lobulated tumorous lesion in the membranous portion of the lower trachea (Figure 2A). The lesion showed extraluminal growth but no definitive signs of invasion to the surrounding areas (Figure 2B). Bronchoscopy demonstrated a wide-based submucosal tumor with prominent vessels on the membranous portion of the lower trachea with protruding irregular-surfaced portions (Figure 3). A biopsy was carefully performed, and based on the specimen obtained, the tumor was diagnosed as a non-malignant schwannoma.

We initially considered whether endotracheal resection would be suitable, owing to the benign nature of the tumor. However, we instead selected a surgical ap-
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Approach, as the tumor had a wide base and extended throughout both the intraluminal and extraluminal tissue, making complete endotracheal resection too difficult.

We performed right posterolateral thoracotomy for the surgical approach. Right posterolateral incision and 4th intercostal thoracotomy were performed, and an intercostal muscle flap was prepared cover of the tracheal anastomosis. The tumor could be easily detected rising from the right side of the membranous portion of the lower trachea; its distal edge was located approximately 3 cm from the carina. The tumor did not invade the surrounding tissues as had been expected. Resection of 3 rings of the trachea and end-to-end anastomosis with interrupted monofilament absorbable sutures were successfully performed with an in-field endotracheal intubation into the distal trachea to the left main bronchus during anastomosis. Intraoperative frozen section showed the margin to be free of tumor.

The immediate postoperative course was uneventful and the patient was discharged on the 10th postoperative day.

The pathologic examination revealed benign schwannoma, which is composed of so-called Antoni type A and B components (Figure 4); and the surgical margin was found to be negative. The tumor proved to exist outside of the tracheal membranous portion on the tissue section. Immunohistochemical staining revealed uniform positivity for S-100 protein.

The bronchoscopic findings 6 months after surgery revealed good healing of the anastomosis site (Figure 5), and at the latest follow-up, the patient was tumor-free.

**DISCUSSION**

While tracheal tumors are relatively rarely reported, a variety of tumors can occur in this region, including primary malignant tumors, secondary malignant tumors, and benign tumors. Among these tumors, primary tracheal schwannomas are especially rare. Grillo and Mathisen,1 in their case series of 198 consecutive primary tracheal tumors, reported only 1 case of schwannoma. In the Japanese literature, only 30 cases of tracheal schwannoma have been reported to date.24

Schwannoma is generally a benign tumor; and, therefore, less invasive procedures should be considered initially. Recently, endoscopic resection for endotracheal tumors has been reported as a feasible treatment option.56 As Kasahara et al. emphasized,7 we believe that pedunculated polypoid lesions without an extraluminal

**Figure 1.** The flow-volume curve pattern suggests an upper airway obstruction (A), which improved after surgical resection of the tracheal tumor (B).
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Figure 2. Chest computed tomography findings. A mass (17 mm in size) protruding from the tracheal membranous portion is observed (A: horizontal section). The lesion showed extraluminal growth (B: sagittal section).

Figure 3. Bronchoscopic examination showed a tracheal polypoid tumor with a glossy and patterned indented surface.

Figure 4. Microscopic examination of the tumor shows characteristic features of benign schwannoma, with spindle cells and palisading nuclei (arrow) (hematoxylin and eosin stain × 40).

component are the most favorable indication for endoscopic resection. However, recent reports have showed some cases with positive resected margins after bronchoscopic resection. Moreover, schwannoma is generally a slow-growing tumor, and long-term postoperative surveillance is needed. Indeed, Horovitz et al. reported a recurrent case of primary intratracheal neurilemoma 12 years after an endoscopic resection.
Under some circumstances, the tumor has a wide base or extends throughout both the intra- and extraluminal tissue, in which case surgery is preferable to achieve complete resection. It is difficult to diagnose precisely the degree of tumor extension, intraluminal or extraluminal before treatment. At the present time, we can only study bronchoscopic findings precisely and put those together with CT or magnetic resonance imaging findings. It was reported that the possibility of extraluminal extension is considerably high.7

However, it should be noted that perioperative complications around the anastomotic site may be fatal. Hence, careful planning, including making detailed arrangements with the anesthetic team, is indispensable. The standard surgical procedure for tracheal tumors is circumferential resection with tracheal reconstruction, whereas partial resection (wedge resection, membranous tracheoplasty, and so on) may be sufficient for a subset of benign tumors. The surgical approach for tracheal tumors is selected according to the site of the tumor and its position in relation to the surrounding tissues. Generally, cervical incision, median sternotomy, or lateral thoracotomy may be selected. For lower tracheal tumors, both median sternotomy and lateral thoracotomy are potential options.19 For the present case, we selected lateral thoracotomy, owing to the visual findings.

Although the tumor was successfully resected, we intend to carefully follow the patient with annual bronchoscopy examinations to determine the presence of any recurrence. The flow-volume curve improved after surgical resection of the tracheal tumor (Figure 1B).

CONCLUSION

We treated a rare case of tracheal schwannoma by partial tracheal resection and end-to-end anastomosis. Because of the benign and slow growing nature of these tumors, bronchoscopic resection is generally thought to be favorable; however, surgery should be considered an alternative procedure for cases in which bronchoscopic resection is not suitable. Precise understanding of the bronchoscopic findings is essential to decide the appropriate treatment.

No potential conflicts of interest are disclosed.

REFERENCES