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## Glomus Tumor of the Trachea Managed by Spiral Tracheoplasty

Authors' Contribution:  
Study Design A  
Data Collection B  
Statistical Analysis C  
Data Interpretation D  
Manuscript Preparation E  
Literature Search F  
Funds Collection G

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**Conflict of interest:** None declared

**Patient:** Female, 58  
**Final Diagnosis:** Glomus tumor of trachea  
**Symptoms:** Hemoptysis  
**Medication:** —  
**Clinical Procedure:** —  
**Specialty:** Otolaryngology

**Objective:** Rare disease

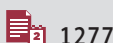
**Background:** Glomus tumors are usually found over the dermis of the extremities, particularly over the subungual region of the fingers, and occurrence in the trachea is an extremely rare event. To date, only 29 cases of tracheal and 2 main bronchus glomus tumors have been reported in the English literature. Our patient is the first ever reported case in Taiwan that was managed by spiral tracheoplasty.

**Case Report:** A 58-year-old woman was admitted to our hospital because of hemoptysis. Computed tomographic (CT) scan revealed a mass over the posterior wall of the trachea. Surgical resection with spiral tracheoplasty was performed due to uncontrolled bleeding and airway compromise. Histopathology and immunostaining confirmed a glomus tumor. Postoperative course was unremarkable and she was discharged in improved condition after 9 days of hospital stay.

**Conclusions:** Although chronic symptom presentation is the rule for tracheal glomus tumors, airway obstruction and bleeding are life-threatening presentations. Histopathological examination and staining are important to differentiate it from hemangiopericytoma or carcinoid tumors. Spiral tracheoplasty after tangential resection may be tried, as this preserves more tracheal tissue, decreases tension, and prevents postoperative leakage at the anastomotic site.

**MeSH Keywords:** Glomus Tumor • Thoracic Surgery • Tracheal Diseases

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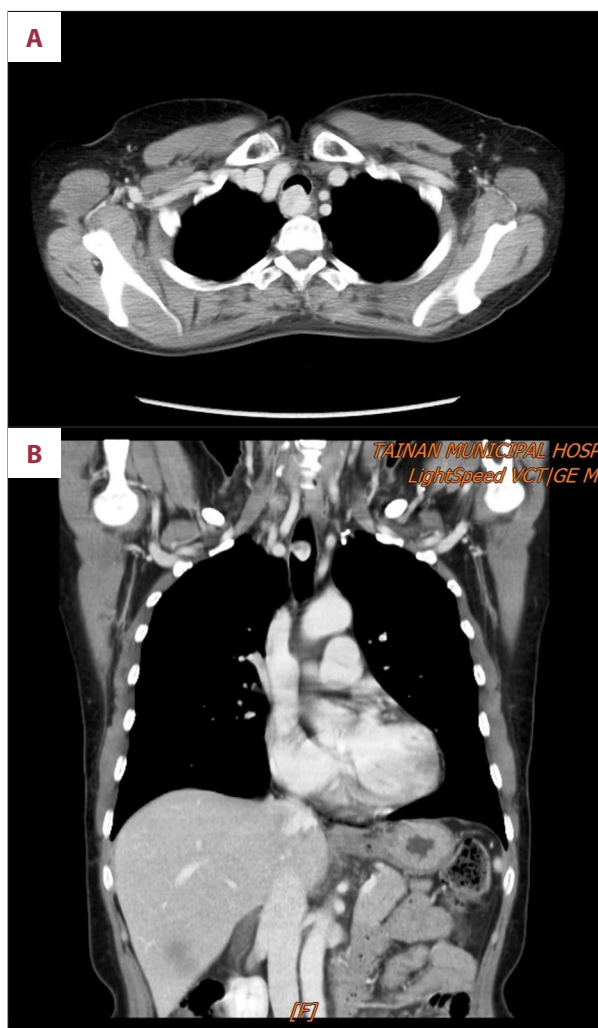
## Background

Glomus tumors are usually found over the subungual region of the fingers of the hand and the lower extremities. Its occurrence in the trachea is extremely rare. To date, there are 31 reported cases of tracheal and proximal main bronchus glomus tumors in the English literature [1–28] since it was first reported by Mason in 1924 [29]. Sleeve resection with primary reconstruction of the trachea is usually the treatment of choice for tracheal glomus tumors. In patients with tangential tracheal lesions, spiral tracheoplasty has never previously been performed, but can be attempted after tangential resection. Our patient is the first reported case to undergo successful spiral tracheoplasty, as pioneered by one of our authors, after resection of this tumor that occurred in a very rare location.

## Case Report

A 58-year-old Taiwanese woman was admitted to our hospital due to hemoptysis. She had no known systemic illnesses in the past and did not smoke cigarettes or consume alcoholic beverages. She denied any weight loss, fever, cough, dyspnea, or epistaxis prior to this incident. A chest CT scan was performed, showing a 2.2×2.2 cm polypoid lesion over the posterior wall of the lower third of the trachea (Figure 1A, 1B).

On examination, the patient was afebrile and breath sounds were clear. Laboratory test results were unremarkable. A bronchoscopic examination was performed, showing a polypoid tumor over the posterior wall of the trachea (Figure 2A). During admission, the patient had another episode of massive hemoptysis, which resulted in hypotension, respiratory distress, and anemia that necessitated a blood transfusion. Our thoracic surgeon was consulted and surgery was performed. Excision of the tracheal tumor via sternotomy was performed (Figure 2B) followed by reconstruction of the trachea with spiral anastomosis. Afterwards, hemostasis was achieved, hemodynamics stabilized, and intraoperative fiberoptic bronchoscopy showed tracheal patency. Hemangiopericytoma was reported on fast-frozen section. However, histopathology showed a hypervascular tumor composed of branching, dilated, thick-walled vascular channels and thin-walled capillary-like vascular spaces, surrounded by lobular arrangements of oval-to-spindle cells, with abundant eosinophilic cytoplasm and centrally-located rounded nuclei. A focal hemangiopericytoma-like pattern interspersed with thin-to-coarse collagenous bundles was also noted (Figure 3A–3B). Immunohistochemical staining was positive for smooth muscle actin (Figure 3C), focally reactive with synaptophysin and negative for cytokeratin, chromogranin A, S-100 protein, and HMB-45. This was later diagnosed as a glomus tumor. Seven days after surgery, fiberoptic bronchoscopy was repeated, showing a patent trachea with mild

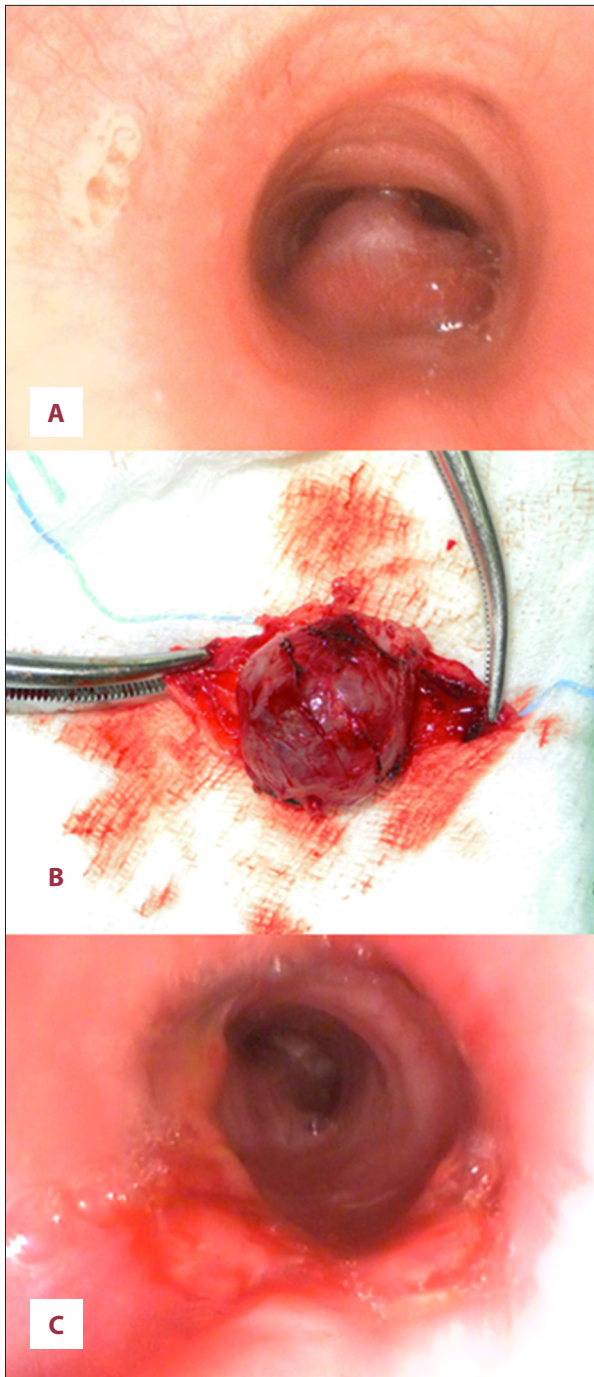


**Figure 1.** (A) Axial CT scan image of the tumor located at the posterior wall of the trachea. (B) Coronal section showing the polypoid tumor over the trachea.

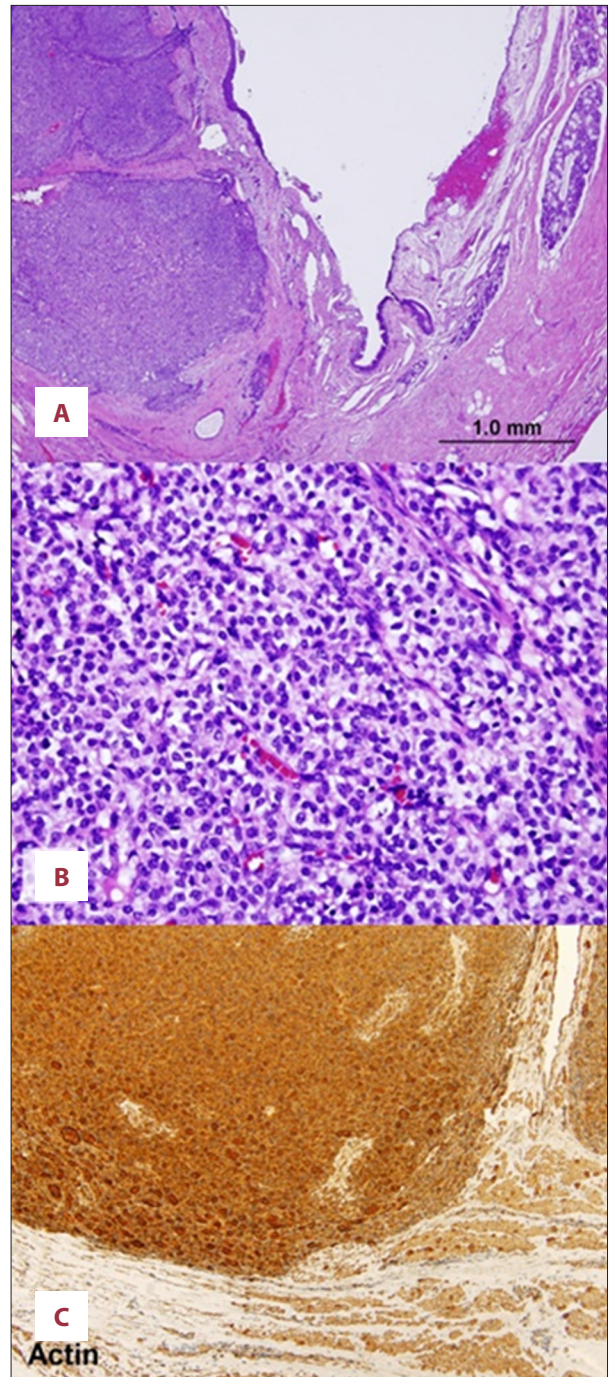
granulation tissue over the oblique anastomotic line and no stenosis (Figure 2C). The patient was discharged the next day. She is currently asymptomatic and no complication or tumor recurrence has occurred in approximately 2 years of follow-up.

## Discussion

Glomus tumors are usually benign and rarely exhibit malignant potential. However, they can cause airway obstruction and bleeding. In 2001, the World Health Organization reclassified these tumors with atypical and malignant features into 4 categories [30]. The diagnosis of malignant glomus tumor is reserved for tumors having: a size of >2 cm and subfascial or visceral location, atypical mitotic figures or marked nuclear atypia, and any level of mitotic activity. Glomus tumors not fulfilling the criteria for malignancy, but having at least 1 atypical



**Figure 2.** (A) Preoperative bronchoscopic image of the tumor almost completely obstructing the trachea. (B) Gross appearance of the excised tumor. (C) Bronchoscopic image performed 7 days postoperatively showing mild granulation tissue over the oblique anastomotic site. The trachea is patent and the axis of the distal anastomotic site is not on the same plane as the proximal segment.



**Figure 3.** (A) Representative low-power magnification of a cut section of the glomus tumor showing a hypervascular tumor composed of branching, dilated, thick-walled, vascular channels and thin-walled, capillary-like vascular spaces (hematoxylin and eosin, 40 $\times$ ). (B) High-power magnification of the tumor showing lobular arrangements of oval- to spindle-shaped cells, with abundant eosinophilic cytoplasm and centrally-located rounded nuclei (hematoxylin and eosin, 400 $\times$ ). (C) Immuno-staining showing tumor cells strongly reactive to actin.



**Table 1.** Tracheal glomus tumor cases from 1950–2014.

No.	Author Reference	Year	Age	Sex	Symptoms	Tumor site	Size (cm)	Treatment	Outcome
1	Hussarek [1]	1950	43	F	Dyspnea, stridor	Upper 3 <sup>rd</sup> post. wall	Bean size	Tracheal resection	Not stated
2	Fabich [2]	1980	63	M	Cough	Lower 3 <sup>rd</sup> post. wall	2.5×2.0×1.0	Sleeve resection	Died of complications on 10 <sup>th</sup> post-op day
3	Warter [3]	1980	69	M	Dyspnea, hemoptysis	Mid-trachea post. wall	2.3×1.5×1.5	Segmental resection	Unremarkable
4	Heard [4]	1982	50	M	Dyspnea, asthma-like symptoms	Lower 3 <sup>rd</sup> post. wall	2.5×1.5×1.0	Sleeve resection	Died of sepsis on 15 <sup>th</sup> post-op day
5	Ito [5]	1988	51	M	Respiratory infection, hemoptysis	Upper 3 <sup>rd</sup> post. wall	1.5×1.2×1.0	Segmental resection	No recurrence at 2 years
6	Sheffield [6]	1988	74	M	Dyspnea, cough	Lower 3 <sup>rd</sup> post. wall	2.2	Endoscopic removal	Unremarkable
7	Kim [7]	1989	54	F	Dyspnea, cough, hemoptysis	Mid-trachea post. wall	1.5×1.2	Segmental resection	No recurrence at 13 months
8	Shin [8]	1990	47	F	Cough, hemoptysis	Lower 3 <sup>rd</sup> post. wall	1.5×1.0×1.0	Wedge resection	Not stated
9	Garcia-Prats [9]	1991	58	M	Dyspnea, cough, hemoptysis	Mid-trachea post. wall	2.5×1.8	Segmental resection	No recurrence at 8 months
10	Haraguchi [10]	1991	61	M	Asymptomatic	Mid-trachea post. wall	1.2	Sleeve resection	Not stated
11	Arapantoni [11]	1995	65	M	Dyspnea, hemoptysis	Lower 3 <sup>rd</sup> post. wall	4.5×3.0	Bronchoscopy with Nd-Yag laser excision	No recurrence at 1 year
12	Koskinen [12]	1998	66	M	Asymptomatic	Lower 3 <sup>rd</sup> post. wall	2.0×3.0	Multiple endoscopic laser ablations and external radiotherapy	Not stated
13	Watanabe [13]	1998	43	M	Hoarseness	Lower 3 <sup>rd</sup> post. wall	2.0×1.6×1.4	Sleeve resection	No recurrence at 20 months
14	Menaissy [14]	2000	34	M	Hemoptysis	Mid-trachea post. wall	2.4×2.1×1.6	Tracheal resection	No recurrence at 4 months
15	Lange [15]	2000	20	M	Dyspnea	Left main bronchus	1.4×1.3×0.6	Bronchial sleeve resection	No recurrence at 9 months
16	Gowan [16]	2001	73	M	Cough, chest pain, dyspnea, hemoptysis	Mid-trachea post. wall	1.6×0.3×0.6	Segmental resection	No recurrence at 6 years
17	Chien [17]	2003	50	F	Cough, dyspnea, hemoptysis	Lower 3 <sup>rd</sup> post. wall	2.5×2.5×2.0	Segmental resection	No recurrence at 1 year
18	Nadrous [18]	2004	39	M	Hemoptysis	Upper 3 <sup>rd</sup> post. wall	2.0×1.5×1.5	Sleeve resection	No recurrence at 3 months
19	Altinok [19]	2006	83	F	Dyspnea, hemoptysis	Upper 3 <sup>rd</sup> post. wall	2.0×1.5×1.2	Partial sleeve resection	No recurrence at 1 year
20	Haver [20]	2008	10	F	Dyspnea	Mid-lower trachea post. wall	1.8×1.3×1.3	Tracheal resection	No recurrence at 2 years

**Table 1 continued.** Tracheal glomus tumor cases from 1950–2014.

No.	Author Reference	Year	Age	Sex	Symptoms	Tumor site	Size (cm)	Treatment	Outcome
21	Colaut [21]	2008	70	M	Dyspnea	Mid-trachea post. wall	2.0×1.0×1.0	Endoscopic resection and Nd-YAG	No recurrence at 2 years
22	Shang [22]	2010	59	M	Chest pain, dyspnea	Lower 3 <sup>rd</sup> post. wall	2.0×1.0×0.5	Endoscopic removal	No recurrence at 1 year
23			22	F	Cough, hemoptysis	Lower 3 <sup>rd</sup> post. wall	1.8×1.5×1.4	Endoscopic removal	No recurrence at 1 year
24	Sakr [23]	2011	66	M	Stridor, cough, dyspnea	Upper 3 <sup>rd</sup> post. wall	1.2×0.8×2.0	Sleeve resection	No recurrence at 21 months
25	Mogi [24]	2011	56	F	Dyspnea, cough	Lower 3 <sup>rd</sup> post. wall	1.3×1.2×1.1	Sleeve resection	No recurrence at 9 months
26	Okereke [25]	2011	58	M	Stridor, dyspnea	Mid-trachea post. wall	1.1	Tracheal resection	No recurrence at 6 months
27	Fan [26]	2013	15	M	Cough, dyspnea, hemoptysis	Mid-trachea post. wall	2.0×2.5	Tracheal resection	No recurrence at 1 year
28			64	M	Asymptomatic	Mid-trachea post. wall	2.6	Tracheal resection	No recurrence at 2 years
29	Choi [27]	2014	52	F	Asymptomatic	Right main bronchus	1.6	Resection of carina and both main bronchi	No recurrence at 3 months
30	Xiong [28]	2014	55	M	Hemoptysis, cough, chest pain	Lower 3 <sup>rd</sup> post. wall	0.5×0.3×0.3	Bronchoscopic cryoablation with brachytherapy	No recurrence at 6 months
31			48	F	Cough, dyspnea	Lower 3 <sup>rd</sup> post. wall	1.2×1.0×0.8	Bronchoscopic cryoablation and argon plasma coagulation	No recurrence at 6 months
32	Wu [current case]	2014	58	F	Hemoptysis	Lower 3 <sup>rd</sup> post. wall	2.2×2.2	Tangential resection with spiral tracheoplasty	No recurrence at 2 years

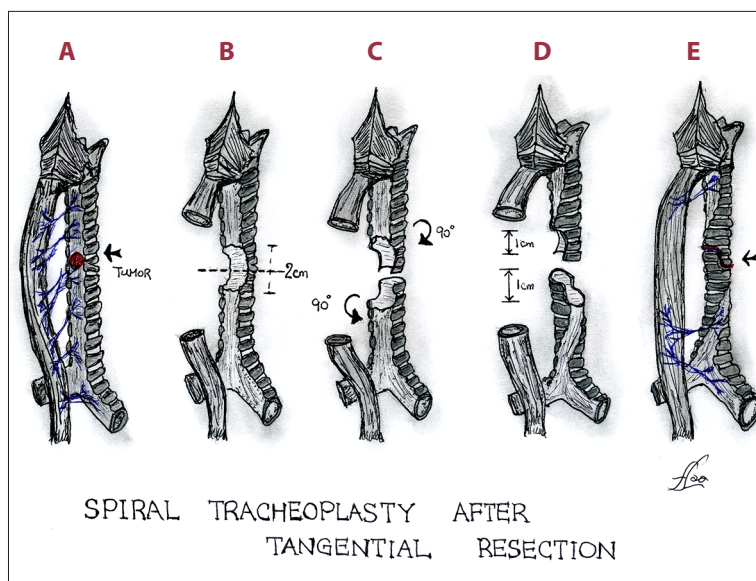
feature other than nuclear pleomorphism, as in this case, are classified as glomus tumor of uncertain malignant potential.

From 1950 to 2014, 29 cases of tracheal and 2 cases of proximal main bronchus glomus tumors have been reported in the English literature. Characteristics of these patients are summarized in Table 1. There were 21 males and 10 females with a mean age of 52±17 years. The youngest was a 10-year-old girl and the oldest was an 83-year-old woman. These tumors occurred anywhere along the length of the trachea and proximal main bronchus, and were all located over the posterior or posterolateral tracheal wall.

Tracheal glomus tumor is sometimes mistaken for a carcinoid tumor, or, as in our case, a hemangiopericytoma. Therefore, immunostaining and careful histopathologic examination should be performed to avoid misdiagnosis. Some would argue for

conservative treatment or watchful waiting for treating this disease since it is benign in 95% of cases. However, complete resection was performed in this case due to recurrent bleeding, acute airway obstruction, and a classification of “uncertain malignant potential”.

Sleeve resection with primary reconstruction of the trachea is the treatment of choice for tracheal glomus tumor. Complete surgical resection is usually curative, but endoscopic intervention may be performed for lesions that are confined to the airway lumen without extension into the airway wall. This can also be done when the tumor is benign, in patients with high surgical risks, or when the patient refuses surgical intervention. Of the 31 cases described in the literature, most underwent surgical resection followed by reconstruction, with only 9 patients receiving endoscopic resection combined with laser ablation.



**Figure 4.** Diagrammatic representation of spiral tracheoplasty after tangential resection of a tracheal tumor. (A) After identifying the tumor, the trachea is separated from the esophagus by about 2 cm to allow for manipulation and exposure of the operative field. (B) This is followed by tangential resection of the tracheal lesion. (C) Both transected tracheal ends are rotated 90 degrees in opposite directions. The proximal end is rotated clockwise and the distal end is rotated counter clockwise. (D) The tracheal ends are then trimmed for irregularities to obtain good apposition. (E) The anastomosis is performed using running sutures.

In 2009, we introduced a technique called spiral tracheoplasty to preserve tracheal tissue while reducing tension at the anastomotic site of the trachea after tangential wall resection (Figure 4). Briefly, the procedure for spiral tracheoplasty consists of 5 steps. 1) After identifying the tumor, the trachea is separated from the esophagus by about 2 cm to allow for manipulation and exposure of the operative field. 2) This is followed by tangential resection of the tracheal lesion instead of a circumferential resection. For example, a 2-cm area of tracheal tissue and tumor was resected in this picture. 3) Both transected tracheal ends are further separated from the esophagus and are rotated 90 degrees in opposite directions. The proximal end is rotated clockwise while the distal end is rotated counter clockwise. 4) The tracheal ends are then trimmed for irregularities to obtain good apposition. In this example, the 1-cm lengths at the proximal and distal ends are positioned next to each other, to obtain a mirror image and are tested for exact fit. 5) Then, the anastomosis is performed using running sutures [31]. Intra- and post-operative fiberoptic bronchoscopy should be performed to check for tracheal patency, bleeding, and optimal anastomosis.

In performing tracheal surgery, it is of utmost importance that the safety of the anastomosis takes precedence over the completeness of the resection. In spiral tracheoplasty, a tangential tracheal wall excision instead of a circumferential resection with end-to-end anastomosis is more advantageous since the resected tracheal tissue is only half of the length removed or lost compared with circumferential resection, preserving and sparing more of the trachea in the process. The tension created by twisting both ends in opposite directions is acceptable,

since a 2-cm or more separation from the esophagus was created prior to the twisting and anastomosis. This important step also frees the esophagus and allows it to shorten or contract a little to accommodate and match the already shortened trachea. This procedure is most suitable for surgery of tangential lesions of the cervicothoracic trachea, and laryngeal release or other types of release of the proximal trachea is unnecessary.

This is the first case wherein a tracheal glomus tumor was managed with this technique, and this is the first time that a bronchoscopic image is available post-procedure. As seen in Figure 2C, the anastomotic line is tangential and oriented to the left, and the axis of the distal anastomotic segment is not in the same plane as the proximal segment.

## Conclusions

Although glomus tumors are mostly benign, tumor location is important because airway compromise and bleeding can be life-threatening. Histopathologic staining is critical to avoid mistaking it for a hemangiopericytoma or a carcinoid tumor. Spiral tracheoplasty may be performed after tangential resection, as it preserves more tracheal tissue, decreases tension, and prevents leakage at the anastomotic site, which is almost always fatal when it occurs.

## Statement

There was no financial support or conflict of interest regarding this manuscript

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