（肺癌．2016;56:12-16）

CASE REPORT

A Case of Glomus Tumor of the Trachea

Yoshimasa Tokunaga[1]; Tatsuo Nakagawa[1]; Masao Saitoh[1]; Takeshi Kondo[1]

ABSTRACT —— ***Background.*** Glomus tumor of the trachea is a rare disease. ***Case.*** We herein describe a 56-year-old man with an asymptomatic glomus tumor of the trachea who underwent tracheal resection and reconstruction. Thirty months after surgery, bronchoscopy and imaging studies revealed no evidence of recurrence. ***Conclusion.*** The clinicopathological features are discussed in this case report.

(*JJLC.* 2016;56:12-16)

KEY WORDS —— Glomus tumor, Trachea, Resection

INTRODUCTION

Glomus tumors of the trachea are rare. To date, only 20 cases in the world have been reported in the literature.[1-4] We herein experienced a surgically treated case of tracheal glomus tumor and described the findings.

CASE REPORT

A 53-year-old man with atrial fibrillation and chronic heart failure was referred to our hospital for further examination of a tracheal lesion detected by chest computed tomography (CT). The patient exhibited no symptoms; however, a chest CT scan revealed a well-defined polypoid lesion measuring 13×18 mm in diameter at the posterior wall of the lower trachea (Figure 1, 2a). FDG-PET did not show a significant uptake at the lesion. Fiberoptic bronchoscopy revealed a tumor lesion covered with intact tracheal mucosa on the right side of the posterior tracheal wall at 4 cm proximal to the carina (Figure 3). A needle biopsy and aspiration were performed,

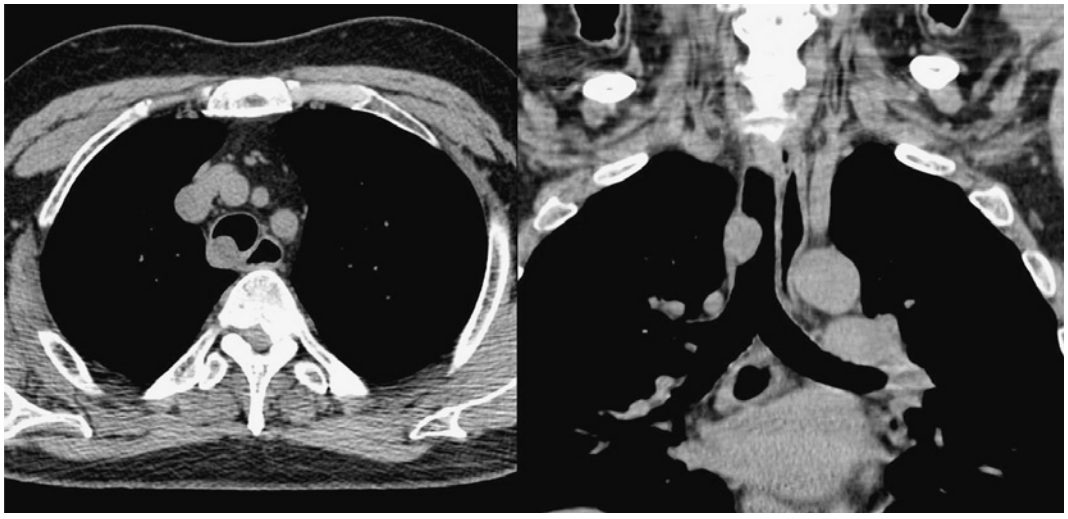

Figure 1. A computed tomography (CT) scan showing a 1.8 cm polypoid lesion arising from the posterior wall of the lower trachea.

[1]Department of Thoracic Surgery, Tenri Hospital, Japan.

Reprints: Yoshimasa Tokunaga, Department of Thoracic Surgery, Tenri Hospital, 200 Mishima, Tenri, Nara 632-8552, Japan (e-mail: tokunaga@med.kagawa-u.ac.jp).

Received May 28, 2015; accepted November 16, 2015.

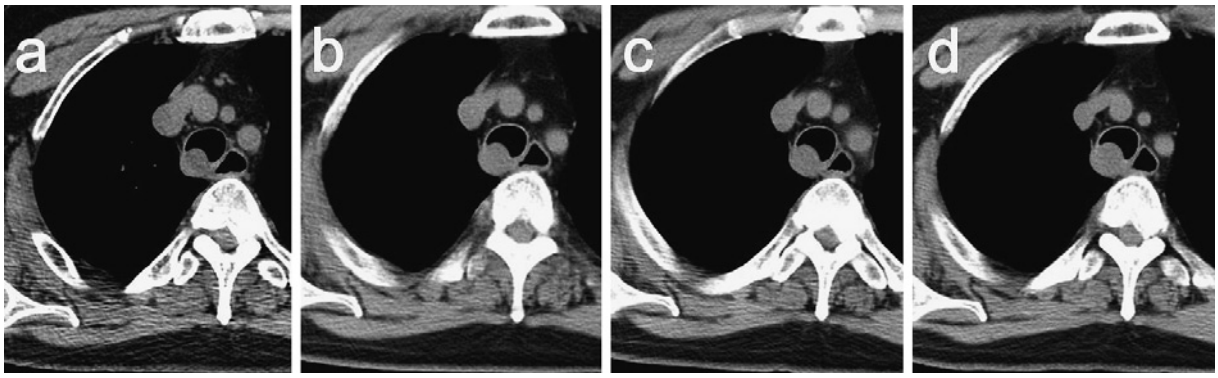

Figure 2. Time series of chest CT: **(a)** first admission; **(b)** 1 year later; **(c)** 2 years later; **(d)** 3 years later.

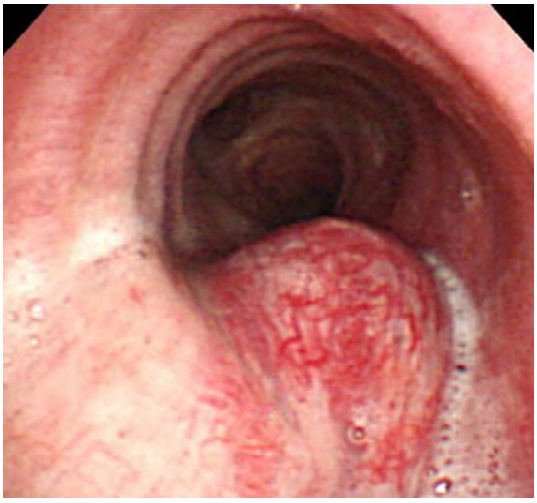

Figure 3. Bronchofiberscopy showing a tumor lesion covered with intact tracheal mucosa on the right side of the posterior tracheal wall at 4 cm proximal to the carina.

however, there was no evidence of malignancy, and a definitive diagnosis could not be determined. The tracheal lesion was suspected to be a benign tumor, such as hamartoma. Due to poorly controlled chronic heart failure, the tumor was followed up by CT every 3 to 6 months for 2 years (Figure 2b, 2c), during which time there was only slight growth. However, chest CT taken 3 years after the initial detection revealed an apparent growth up to 18 × 21 mm in diameter (Figure 2d). A bronchoscopic needle biopsy was performed at this time, but a pathological diagnosis could not be reached. Because the patient's chronic heart failure improved during this 3-year period, surgical resection was performed in March 2011 for the diagnosis and treatment. Median sternotomy and a collar skin incision were performed to enter the mediastinum. The trachea was exposed and mobilized, and the tumor was detected in the lower one-third of the trachea with no invasion to the surrounding tissue. The trachea was transected below the level of the tumor followed by the division of the proximal margin of the tumor. A total of 4 rings of tracheal cartilage were resected, including an additional cartilage resection at the proximal end, and the resected specimen was sent for an immediate pathological examination. The intraoperative diagnosis was carcinoid of the trachea, and microscopically negative margins were obtained at both proximal and distant ends. The head and neck were flexed to remove tension, and the trachea was reconstructed as follows: the posterior wall was first anastomosed using continuous 4-0 monofilament sutures, then the anterolateral wall was anastomosed using individual 4-0 monofilament sutures. During anastomosis of the trachea, ventilation was secured using a sterile oral endotracheal tube passed across the operation field or a high-frequency jet ventilator. The anastomosis was checked for air leakage and covered with the thymus.

The resected trachea measured 30 mm in length, and the tumor was located on the posterior wall of the trachea. The maximum length of the tumor measured 20 mm and extended through the tracheal wall into the extratracheal surrounding tissue, but remained covered with intact epithelium and was well circumscribed without invasion to the surrounding tissue or surgical margin. Microscopically, the tumor consisted of cells with small, round nuclei arranged in sheets and stroma with abundant blood vessels (Figure 4a). The histology of the tumor exhibited a hemangiopericytoma-like pattern, and mitosis was not apparent in the tumor cells. Immu-

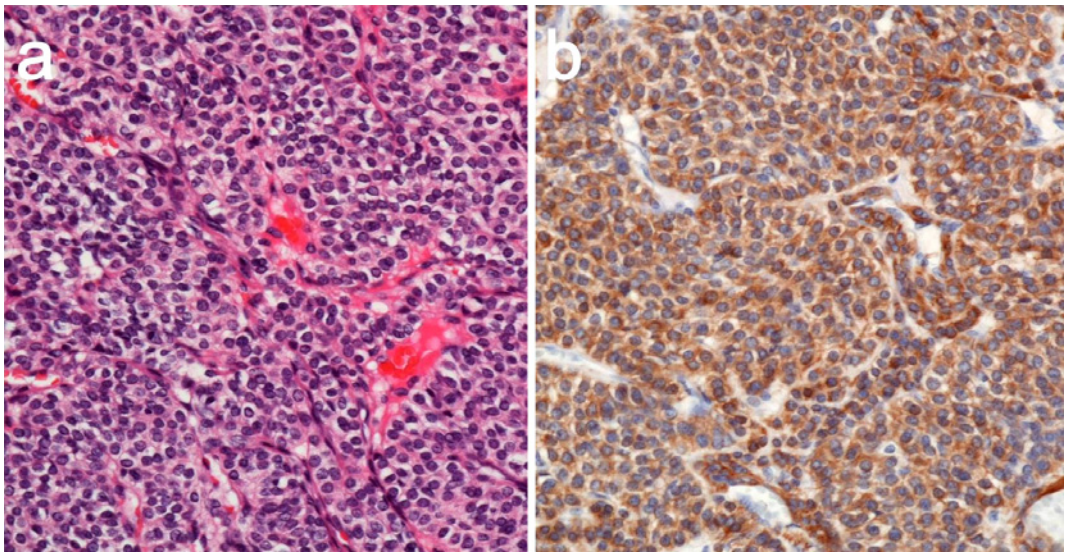

Figure 4. Microscopically, the tumor consisted of cells with small, round nuclei arranged in sheets and stroma with abundant blood vessels **(a**, H&E stain**)**, and tumor cells were immunoreactive for α-actin **(b**, immunostaining for α-actin**)**.

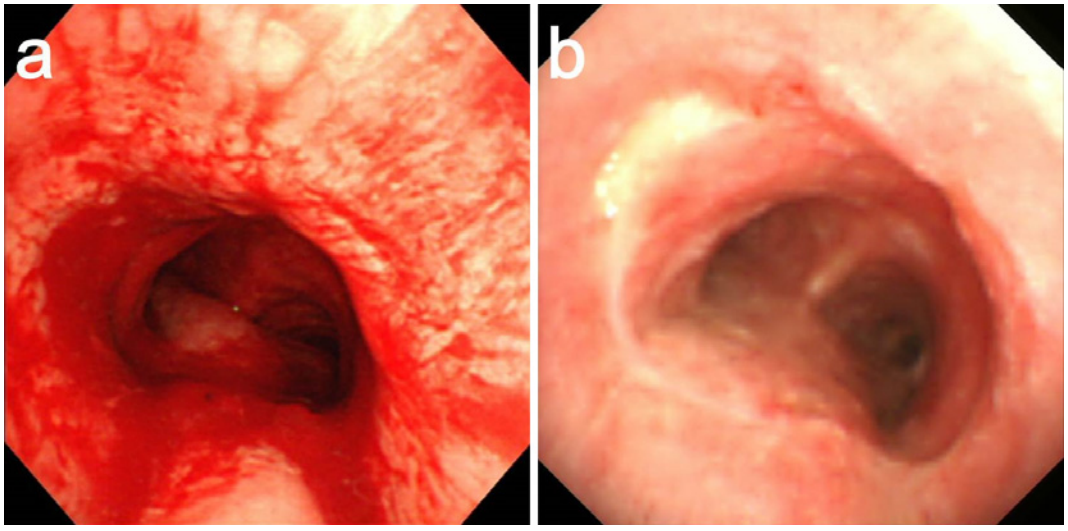

Figure 5. Time series of bronchofiberscopy: **(a)** postoperative day 7; **(b)** postoperative day 80.

nohistochemical staining showed that the tumor cells were positive for α-actin (Figure 4b) and partially positive for synaptophysin, but were negative for CD56 and chromogranin A. The histopathological features of the tumor represented a glomus tumor rather than a carcinoid tumor, which was initially suspected during the immediate intraoperative pathological examination.

The postoperative course was uneventful and the patient was discharged on the 8th postoperative day. Anastomotic failure or tracheal stenosis was not detected 7 days (Figure 5a) and 80 days (Figure 5b) after surgery. There is no evidence of tumor recurrence 30 months after surgery.

DISCUSSION

Glomus tumors are uncommon, benign neoplasms that are found particularly in the subungual region of the finger. The tumor cells are considered to arise from the glomus body, which consists of an arteriovenous shunt surrounded by a capsule of connective tissue in the dermis layer of the skin and involved in body temperature regulation. Glomus tumors share several morphological

Table 1. Reported Cases of Tracheal Glomus Tumors

Authors	Year	Age (yr) Sex	Clinical Features	Location in Trachea	Size (cm)	Treatment	Follow-up
Hussarek and Rieder	1950	43, F	Dyspnea	Subglottic	Bean-sized	Resection	
Fabich and Hafez	1980	63, M	Dyspnea	Above bifurcation	2.5×2×1	Tracheal resection	
Heard et al.	1982	50, M	Asthma-like	Lower third	2.5×1.5×1	Tracheal resection	Sepsis-death
Ito et al.	1988	51, M	Hemoptysis	Upper third	1.5×1.2×1	Tracheal resection	2 yr, no recurrence
Kim et al.	1989	54, F	Asthma-like	Mid-trachea	1.5×1.2	Tracheal resection	13 mo, no recurrence
Shin et al.	1990	47, F	Hemoptysis	Above carina	1.5×1×1	Tracheal resection	
García-Prats et al.	1991	58, M	Cough, dyspnea	Mid-trachea	2.5×1.8	Tracheal resection	8 mo, no recurrence
Haraguchi et al.	1991	61, M	Asymptomatic	Mid-trachea	1.2	Tracheal resection	
Arapantoni-Dadioti et al.	1995	65, M	Hemoptysis, dyspnea	Lower third	Fragments	Nd:YAG	1 yr, no recurrence
Watanabe et al.	1998	43, M	Hoarseness	Lower third	2.0×1.6×1.4	Tracheal resection	20 mo, no recurrence
Koskinen et al.	1998	66, M	Asymptomatic	Lower third		Nd:YAG; external RT	
Menaissy et al.	2000	34, M	Hemoptysis	Mid-trachea	2.4×2.1×1.6	Tracheal resection	4 mo, no recurrence
Gowan et al.	2000	73, M	Dyspnea, hemoptysis	Mid-trachea	1.6×1.3×0.6	Tracheal resection	6 yr, no recurrence
Lange et al.	2000	20, M	Dyspnea	Left main bronchus	1.5×1.0×0.4	Sleeve resection	
Chien et al.	2002	50, F	Dyspnea, hemoptysis	Lower third	2.5×2.5×2.0	Tracheal resection	12 mo, no recurrence
Nadrous et al.	2004	39, M	Hemoptysis	Upper trachea	2.0×1.5×1.5	Tracheal resection	
Colaut et al.	2008	70, M	Dyspnea, wheezing	Mid-trachea	2×1×1	Nd:YAG	24 mo, airway patency, residual disease
Shang et al.	2010	59, M	Chest pain	Lower third	2×1×0.5	Snaring and APC	12 mo, no recurrence
		22, F	Cough, hemoptysis	Lower third	1.8×1.5×1.4	Snaring and APC	12 mo, no recurrence
Mogi et al	2011	56, F	Dyspnea, wheezing	Above carina	1.3×1.2×1.1	Tracheal resection	9 mo, no recurrence
Present case	2012	56, M	Asymptomatic	Lower third	2	Tracheal resection	30 mo, no recurrence

Modified and updated from Colaut F et al. J Thorac Oncol. 2008;3:1065-7.

features with carcinoid tumors. Indeed, Lange et al. reported that 3 cases of 13 glomus tumors were initially diagnosed as carcinoid tumors, after immunohistochemical staining or electron microscopy.[5] Similarly, the resected specimen of the present case was initially diagnosed as a carcinoid tumor. However, staining for smooth muscle actin, and negativity for CD56, chromogranin A, and synaptophysin, supported the diagnosis of a glomus tumor rather than a carcinoid tumor.

Glomus tumors were first described by Hoyer in 1877, while the first complete clinical description was given by Masson in 1924.[6] They can be found in any part of the body, but the trachea is a very unusual site for this tumor, along with the stomach, bone, heart, mediastinum, vagina and lung.[7] To the best of our knowledge, only 21 cases of glomus tumors of the trachea, including the present case, have been reported in the literature (Table 1). These patients had an average age of 51.4 years (range 20 to 73), with a male-to-female ratio of 5:2. Most were symptomatic, presenting with dyspnea, cough, and hemoptysis. All tumors arose from the posterior wall of the trachea, and most were in the lower one-

third of the trachea. They had an average maximal diameter of 1.9 cm (range 1.2-2.5 cm). Radical treatment for the disease includes segmental resection of the trachea with primary reconstruction. In 5 reports, bronchoscopic resection of the tracheal tumor was performed to relieve airway obstruction without surgical resection.[2,3,8,9] Glomus tumors are considered to have a good prognosis due to a relatively benign histology. However, a case of local recurrence was reported.[9] The actual prognosis remains unclear due to the small number of cases and brief follow-up study. In some reported cases,[1,10] the tumor extended outside the tracheal wall. In the present case, the pathological findings of vascular invasion or destruction of normal tracheal structure, which indicates the malignancy potential, was partially observed, although the morphological features of each cell did not suggest malignancy. Regarding these issues, whenever the diagnosis of glomus tumor is obtained, radical resection of the trachea should be considered rather than conservative therapies, such as bronchoscopic resection or radiotherapy, except in high-risk patients.

本論文内容に関連する著者の利益相反：なし

The gist of the present paper was presented at The 52nd Annual Meeting of the Japan Lung Cancer Society.

REFERENCES

1. Menaissy YM, Gal AA, Mansour KA. Glomus tumor of the trachea. *Ann Thorac Surg.* 2000;70:295-297.
2. Colaut F, Toniolo L, Scapinello A, Pozzobon M. Tracheal glomus tumor successfully resected with rigid bronchoscopy: a case report. *J Thorac Oncol.* 2008;3:1065-1067.
3. Shang Y, Huang Y, Huang HD, Bai C, Dong YC, Zhao LJ, et al. Removal of glomus tumor in the lower tracheal segment with a flexible bronchoscope: report of two cases. *Intern Med.* 2010;49:865-869.
4. Mogi A, Kosaka T, Yamaki E, Tanaka S, Kuwano H. Successful resection of a glomus tumor of the trachea. *Gen Thorac Cardiovasc Surg.* 2011;59:815-818.
5. Lange TH, Magee MJ, Boley TM, Bell SW, Hazelrigg SR. Tracheobronchial glomus tumor. *Ann Thorac Surg.* 2000;70:292-295.
6. Masson P. Le glomus neuromyoarterial des regions tactiles et ses tumours. *Lyon Chir.* 1924;21:257-280.
7. Shin DH, Park SS, Lee JH, Park MH, Lee JD. Oncocytic glomus tumor of the trachea. *Chest.* 1990;98:1021-1023.
8. Arapantoni-Dadioti P, Panayiotides J, Fatsis M, Antypas G. Tracheal glomus tumour. *Respiration.* 1995;62:160-162.
9. Koskinen SK, Niemi PT, Ekfors TO, Sipilä J, Valavaara R, Dean PB. Glomus tumor of the trachea. *Eur Radiol.* 1998;8:364-366.
10. García-Prats MD, Sotelo-Rodríguez MT, Ballestín C, Martínez-González MA, Roca R, Alfaro J, et al. Glomus tumour of the trachea: report of a case with microscopic, ultrastructural and immunohistochemical examination and review of the literature. *Histopathology.* 1991;19:459-464.