

Glomangiomyoma of the Trachea

Sam Hyun Baek, M.D.*, Dong Myung Huh, M.D.**, Jun Ho Park, M.D.*, Eun Kyoung Kwak, M.D.***, Byung Ho Kim, M.D.**, Won Kyung Han, M.D.**

A glomus tumor is an uncommon soft tissue tumor that is most commonly found in the subungual area and a glomus originating in the trachea is extremely rare. Histologically and ultrastructurally, these tumors have been divided into three subtypes: classic glomus tumors, glomangiomas, and glomangiomyomas. Glomangiomyomas account for less than 10% of all glomus tumors and are the least common type. We report a case of a 54-year-old man with glomangiomyoma of the trachea who presented with stridor. We treated the tumor by segmental resection and primary repair via a transcervical approach.

Key words: 1. Tracheal neoplasms
2. Trachea
3. Glomangiomyoma

CASE REPORT

A 54-year-old man presented with a 3-month history of cough and shortness of breath during exercise. On physical examination, he had an audible expiratory stridor. The chest X-ray was normal. Computed tomography (CT) of the neck and chest revealed a 1.5 cm polypoid tumor arising from the posterior wall of the upper thoracic trachea (Fig. 1). A flexible bronchoscopy showed a double cone shaped, polypoid tumor arising from the posterior wall of the trachea, 5 cm distal to the vocal cords, and 7 cm proximal to the carina (Fig. 2). Based on the biopsy specimens, the mass was diagnosed as a glomus tumor. To check for an extension of the tumor into the surrounding tissue, an endoscopic ultrasonic exam was performed, and it showed that the tumor was confined to the trachea. Since the tumor was located in the upper trachea,

we decided to approach the tumor through a cervical collar incision. After exposure of the trachea, the location of the tumor was identified between the 5th and 6th tracheal rings using flexible bronchoscopy. We divided the trachea one cartilage ring below the tumor and a sterile laryngectomy tube was inserted into the distal trachea. A proximal tracheal division was made one cartilage ring above the tumor. A frozen section analysis confirmed negative margins. Anastomotic sutures were placed using interrupted 4-0 Vicryl sutures (Ethicon, Somerville, NJ).

The surgical specimen was 3 cm in length with a luminal diameter of 2.3 cm. There was a 1.3×1.2 cm blue-red protruding nodule on the mucosal surface of the trachea (Fig. 3). The tumor did not show an infiltrative growth into the tracheal cartilage. At the microscopic level, the tumor showed glomus cells, vascular structures, and smooth muscle tissue

*Department of Otolaryngology, Daegu-Fatima Hospital

**Department of Thoracic and Cardiovascular Surgery, Daegu-Fatima Hospital

***Department of Pathology, Daegu-Fatima Hospital

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Corresponding author: Dong Myung Huh, Department of Thoracic and Cardiovascular Surgery, Daegu-Fatima Hospital, 576-31, Sinam-dong, Dong-gu, Daegu 701-819, Korea
(Tel) 82-53-940-7250 (Fax) 82-53-940-7255 (E-mail) dmhuh@naver.Com

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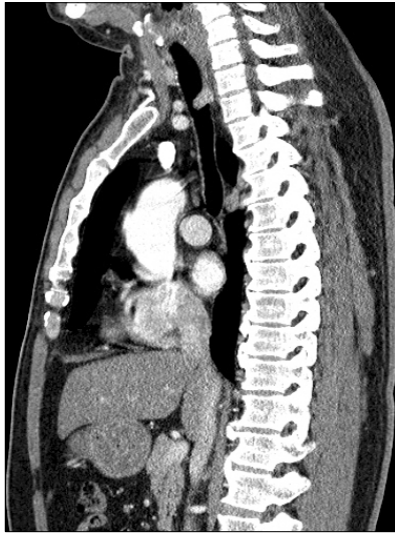


Fig. 1. Computed tomographic scan of the chest shows a well-enhanced, polypoid tumor arising from the posterior wall of the upper thoracic trachea.

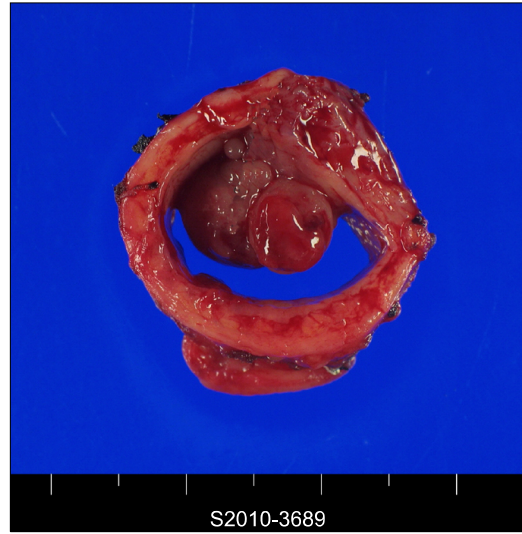


Fig. 3. Resected tracheal specimen shows a protruding polypoid mass arising from the posterior wall of the trachea.

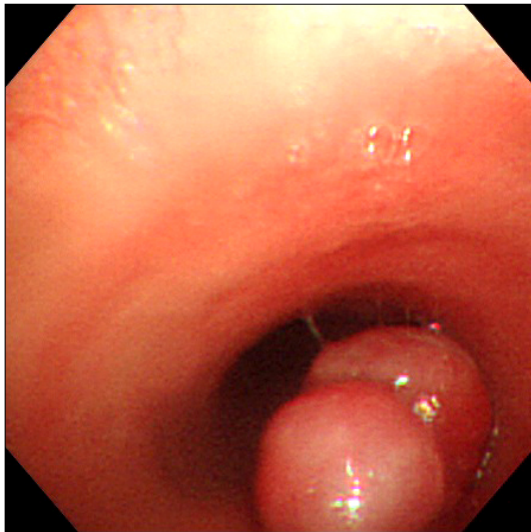


Fig. 2. Bronchoscopic view demonstrates an intraluminally protruding double cone-shaped tumor.

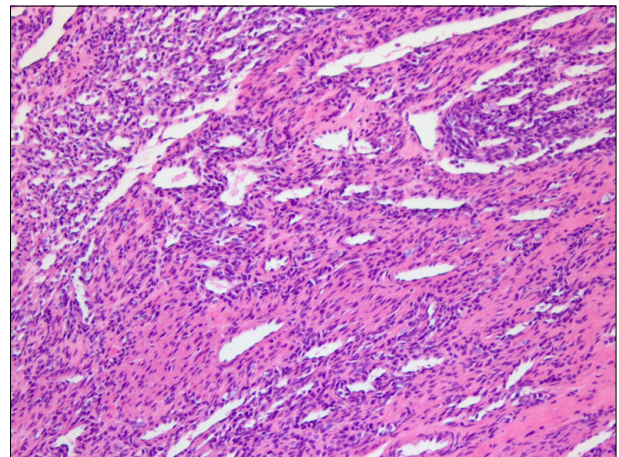


Fig. 4. The tumor is composed of mostly small round to oval cells and is rich in blood vessels and smooth muscle tissue (H&E).

(Fig. 4). The glomus cells had undergone a transition to elongated, mature smooth muscle cells in some foci (Fig. 5); this feature differentiated this tumor from ordinary glomus tumors. The tumor cells showed positive reactions for smooth muscle actin and CD34, but negative reactions for S-100 and cytokeratin on immunohistochemical staining. These results led to

the diagnosis of glomangiomyoma.

Bronchoscopy and a chest CT were performed 3 months after the operation.

They showed well anastomosed tracheal mucosa and a good tracheal air column (Fig. 6). The patient was asymptomatic for 24 months after surgery.

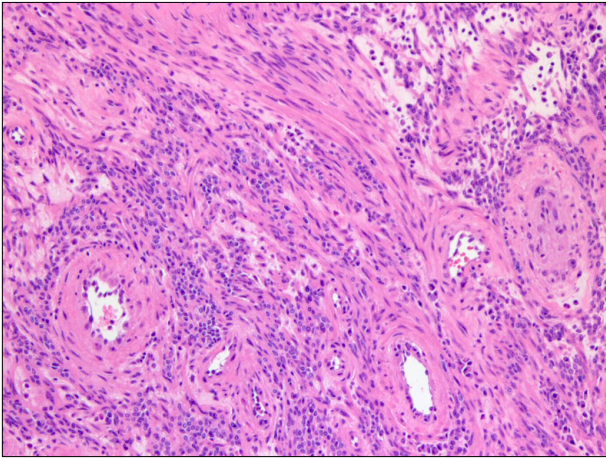


Fig. 5. There are several foci of gradual transition from glomus cells to elongated, smooth muscle cells (H&E).

DISCUSSION

Glomus tumors are uncommon neoplasms, which are typically seen in the deep dermis or subcutis of the extremities, most commonly in the subungual region of the finger. However, these tumors also occur at sites where normal glomus bodies may be sparse or even absent, such as the patella, chest wall, bone, stomach, colon, nerve, eyelid, nose, mediastinum, and trachea [1].

Histologically and ultrastructurally, the tumor cells have varying proportions of glomus cells, vascular structures, and smooth muscle tissue. According to the relative proportions, they have been divided into three subtypes: glomus tumor proper, glomangioma, and glomangiomyoma. Classic glomus tumors account for about three-fourths of all glomus tumors. Glomangiomas constitute about one-fifth of all glomus tumors. Glomangiomyomas account for less than 10% of all glomus tumors and are the least frequent type. The overall pattern of glomangiomyomas may be identical to that of an ordinary glomus tumor or a glomangioma. However, there is a gradual transition from glomus cells to elongated, mature smooth muscle cells [1].

For the tumor presented here, the initial bronchoscopic biopsy specimen diagnosed the tumor as a glomus tumor. However, the resected tumor had features of a glomangiomyoma such as glomus cells, vascular structures, and smooth muscle tissue, and the glomus cells undergo gradual transition



Fig. 6. Postoperative computed tomographic scan of the neck reveals a normal tracheal air column.

to smooth muscle cells in some foci. These features suggest the diagnosis of a glomangiomyoma. These findings are supported by various immunohistochemical studies in which glomus tumors show positive reactions for smooth muscle actin and CD34, and negative reactions for S-100 and cytokeratin.

Since Masson first described this tumor in 1924, sporadic reports of glomus tumors of varying types and locations have been reported in the literature [2]. While the trachea is a very rare site for this tumor, segmental resection of the trachea with primary reconstruction is the treatment of choice for these tumors [3,4]. For some patients, bronchoscopic resection of the tumor or tracheal stenting was useful in relieving airway obstruction before operation. Bronchoscopic resection and laser surgery should be reserved for high-risk patients, as local recurrence may occur [5,6].

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