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【 CASE REPORT 】

Successful Treatment of Life-threatening Tracheal Stenosis Caused by Malignancy with a Self-expanding Hybrid Stent

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Abstract:
Tracheal stenosis caused by malignancy is a life-threatening complication. We performed stent therapy in two patients using the AERO™ stent, launched in late 2016 in Japan. One patient presented with stenosis of the trachea due to adenoid cystic carcinoma and the other with stenosis of the trachea due to esophageal cancer. Both patients showed improved symptoms, and no complications were identified. This is the first report of a favorable outcome with the use of this hybrid stent in Japan, and the findings suggest that insertion of the AERO hybrid stent is an effective way to improve patients' quality of life.

Key words: tracheal stenosis, adenoid cystic carcinoma, esophageal cancer, hybrid stent, AERO™ stent

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Introduction

Tracheal stenosis due to malignancies such as lung cancer or esophageal cancer is a life-threatening complication, and its diagnosis is usually difficult. In the past, silicone and metallic stents have been used for the treatment of such a clinical condition. Although these stents are very effective, silicone stents are difficult to secure with a tracheal lesion, and metallic stents have the disadvantage of tumor extension into the stent.

The AERO™ stent (Merit Medical Systems, South Jordan, USA), which was first released in late 2016 in Japan, is a self-expanding hybrid stent that combines the features of metal and silicone stents with a completely covered nitinol framework. We herein report our experience with two cases, wherein the AERO stent was successfully used to treat tracheal stenosis due to malignancy.

Case Reports

Case 1

A 57-year-old man was admitted to Miyazaki Prefectural Miyazaki Hospital with dyspnea. The flow-volume curve showed a central airway obstruction pattern. There was decreased radiolucency in the middle part of the trachea on chest X-ray (Fig. 1a). Chest computed tomography (CT) revealed a tumor arising from the posterior wall of the middle portion of the trachea and stenosis with wall thickening in the middle portion of the trachea. The most restricted part of the trachea was approximately 7 mm in diameter and 40 mm in length. In addition, part of the border between the trachea and esophagus was indistinct (Fig. 1b and c), and numerous nodular shadows were present in both lungs.

Because we judged the patient to be at high risk of suffocation, we decided to perform tracheal stenting. We selected the AERO stent because it can be removed from the lesion site after treatments such as radiation and because of its solidity in supporting tracheal lesions. However, we decided to perform the procedure in two stages because an AERO stent was not immediately available. First, we cauterized the tumor by argon plasma coagulation and balloon dilation of the stenosis using rigid bronchoscopy under general anesthesia. As a result, the airway stenosis improved by approximately 70%. Second, because of the possibility of re-stenosis due to edema after radiotherapy, we implanted an AERO stent (16× 60 mm, over-the-wire delivery system) under general anesthesia using rigid bronchoscopy (Fig. 2) 5 days after the first procedure. We determined the stent size by measuring the diameter of the narrow segment on CT and directly measuring the narrow segment with an AEROSIZER tra-

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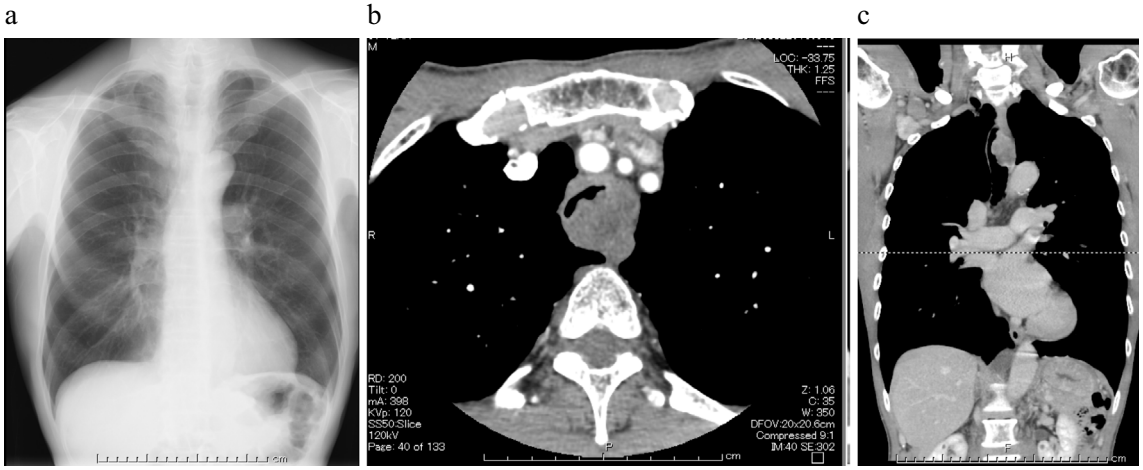

Figure 1. **Chest X-ray and computed tomography (CT) in Case 1. a: There is decreased radiolucency in the middle part of the trachea. b: A chest CT image demonstrates a tumor arising from the posterior wall of the middle portion of the trachea and stenosis with wall thickening of the middle part of the trachea. The most restricted part of the trachea is approximately 7 mm in diameter and 40 mm in length. c: Frontal section view on chest CT.**

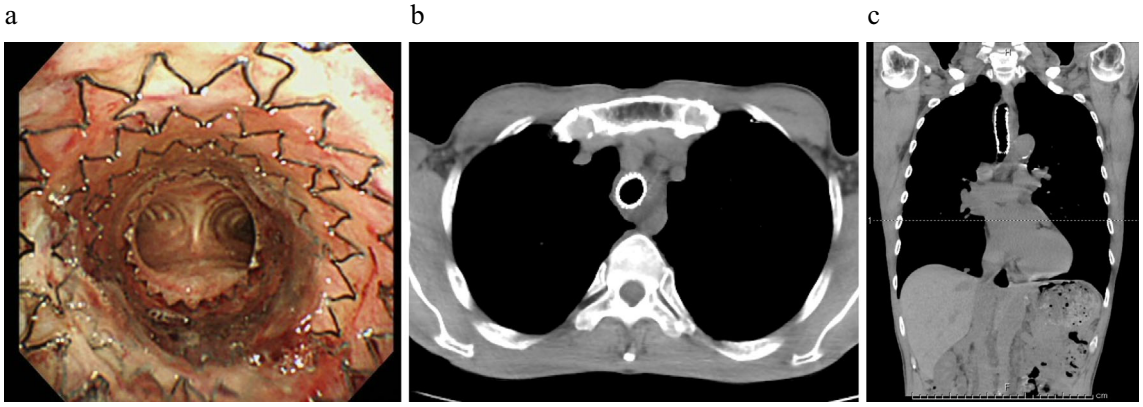

Figure 2. **Endoscopic view and chest computed tomography (CT) after stent placement. a: The stent has expanded sufficiently in the trachea. At the distal end of the stent, the carina is visible. b: A chest CT image shows that the stent has expanded adequately, and the airway is patent. c: Frontal section view on chest CT.**

cheobronchial stent-sizing system. The AERO stent implantation resulted in the dilation of the stenotic trachea; consequently, the patient's sensation of dyspnea disappeared, and the pulmonary function test results improved (Fig. 3).

Adenoid cystic carcinoma (ACC) was diagnosed on an examination of biopsy samples obtained from the lesion, and radiotherapy was performed for local control after insertion of the stent. Further, the patient was diagnosed with stage IV ACC based on examination results of biopsy samples of a small peripheral nodule. There were no signs of stent migration or other complications. Taken together, we decided not to remove the AERO stent in consideration of the patient's poor prognosis. The patient was discharged from our hospital and referred to another hospital for palliative care. Six months after stent placement, the patient was still alive, and neither stent migration nor bleeding had been reported.

Case 2

A 64-year-old man was admitted to our hospital with a productive cough and hoarseness. Chest X-ray revealed an hourglass-like stenosis in the middle part of the trachea (Fig. 4a). Chest CT revealed a mass lesion mainly involving the cervical esophagus, and esophageal cancer was suspected. The esophageal mass extended around the upper part of the trachea, and several enlarged mediastinal lymph nodes were present. Together, the esophageal mass and mediastinal lymph nodes compressed the trachea. The most restricted part of the trachea was approximately 4 mm in diameter and 60 mm in length (Fig. 4b and c). The flow-volume curve showed a central airway-obstruction pattern.

Because the patient was in danger of suffocation, we decided to conduct tracheal AERO stent placement. As the patient had spondylosis, we decided to avoid rigid broncho-

		Before stenting	After stenting
VC	(L)	4.14	4.66
%VC	(%)	93.2	106.2
FVC	(L)	3.74	4.73
%FVC	(%)	86.6	110.8
FEV1.0	(L)	1.26	**3.73**
FEV1.0%	(%)	33.69	**78.86**

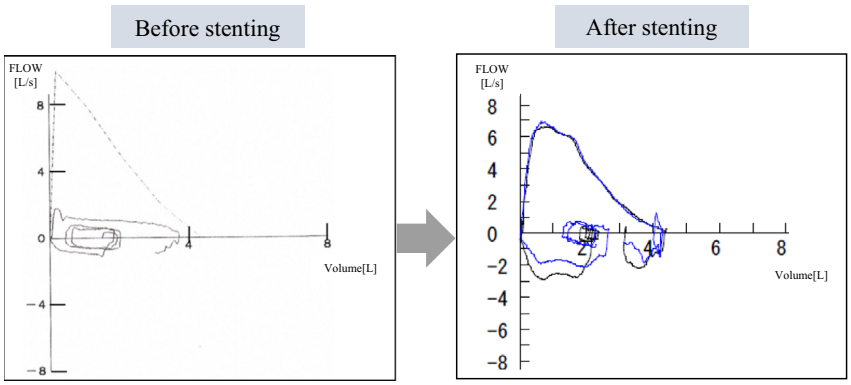

Figure 3. **Pulmonary function test of case 1.**

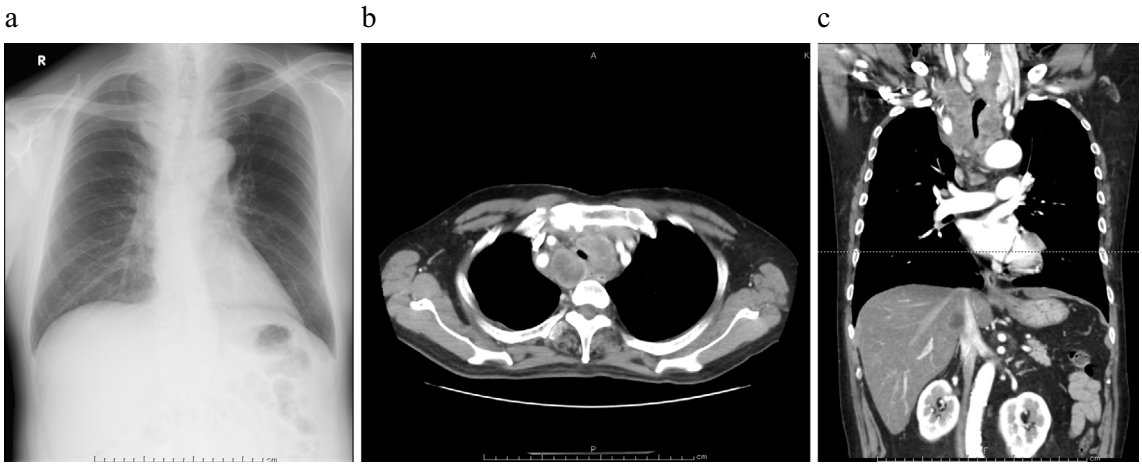

Figure 4. **Chest X-ray and computed tomography (CT) in Case 2. a: A chest X-ray reveals hourglass-like stenosis in the middle part of the trachea. b: A contrast-enhanced chest CT image shows that the narrowest part of the trachea is approximately 4 mm in diameter and that the tumor has progressed to surround the upper part of the trachea. Several enlarged mediastinal lymph nodes are present. c: Frontal section view.**

scopy and to maintain the airway with an 8.5-mm spiral tube. In addition, we performed tracheal stenting after conducted veno-arterial extracorporeal membrane oxygenation (VA ECMO) because we were concerned that the procedure would take longer than with rigid bronchoscopy.

The lesion was present in the upper part of the trachea, and we selected the AERO stent because of its reported stability in such lesions. Because the stenosis was caused by extrinsic pressure, we inserted the AERO stent (16×80 mm, over-the-wire delivery system) (Fig. 5a and b) after balloon expansion. After stent placement, the patient's dyspnea disappeared, and the pulmonary function test results improved (Fig. 6).

Upper gastrointestinal endoscopy revealed esophageal stenosis from the esophageal orifice to 25 cm from the incisor line. Squamous cell carcinoma (SCC) was diagnosed based on an examination of a biopsy sample obtained from the site.

After stent placement, we regularly administered a short-acting beta agonist via inhalation and performed bronchial lavage with the aid of a bronchoscope. The overall status of the patient was good, so he was transferred to another hospital for esophageal cancer treatment. One month after stent placement, there were no reports of stent migration or bleed-

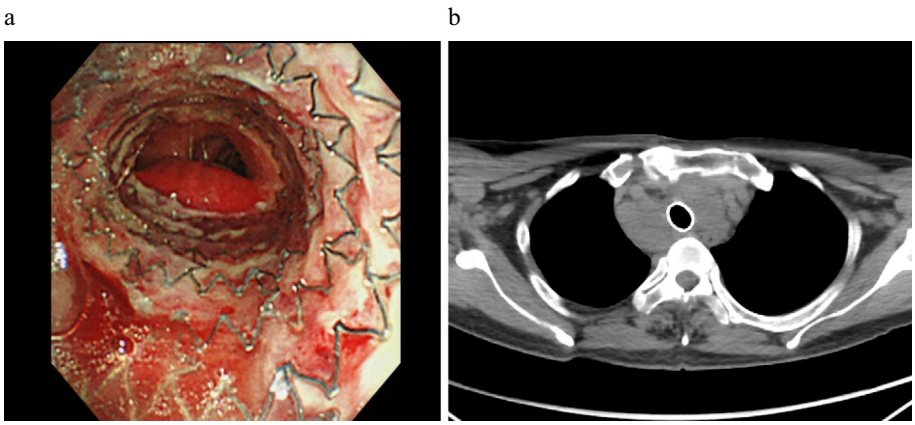

Figure 5. **Endoscopic view and chest computed tomography (CT) after stent placement. a: The stent has expanded sufficiently in the trachea. The carina is visible at the distal end of the stent. b: A chest CT image reveals that the stent has adequately expanded at the narrowest part of the trachea, and the airway is patent.**

		Before stenting	After stenting
VC	(L)	3.61	3.27
%VC	(%)	88.5	80.1
FVC	(L)	3.50	3.27
%FVC	(%)	88.2	82.4
FEV1.0	(L)	1.94	**2.75**
FEV1.0%	(%)	59.1	**83.8**

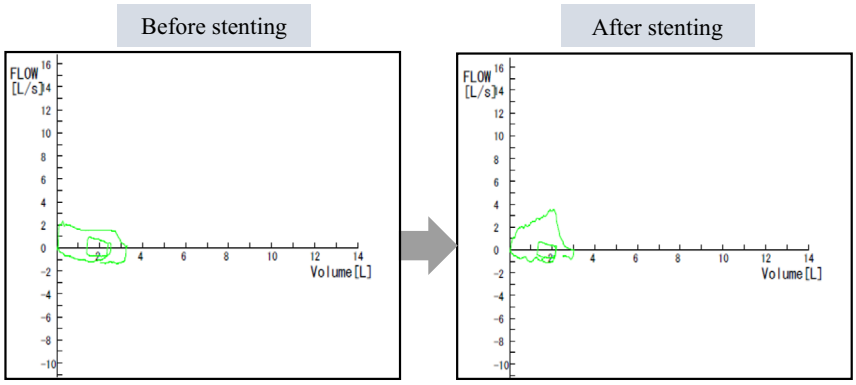

Figure 6. **Pulmonary function test of case 2.**

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Discussion

We reported our experiences with two cases, in which insertion of an AERO stent was effective for tracheal stenosis due to a malignant tumor. To our knowledge, this is the first report of an effective outcome after treatment with the AERO stent in Japan.

Neoplasms of the trachea are uncommon. The incidence of primary tracheal cancer is estimated to be 1 in every 1,000,000 people per year (1). The majority (90%) of tracheal tumors in adults are malignant, with SCC and adenoid ACC being the most common, and metastatic tracheal lesions are much more common than primary tracheal tumors. Direct invasion of the trachea and main bronchi can occur with thyroid, laryngeal, and esophageal cancers (2, 3). Considering the rarity of these neoplasms and their nonspecific clinical presentations, the diagnosis is often delayed by months or even years (4). In addition, it is hard to detect tracheal stenosis on chest X-ray, and patients do not present with symptoms of stenosis, such as dyspnea, stridor, and wheezing, until the stenosis progresses. Therefore when a patient complains of refractory cough and dyspnea, we should consider tracheal stenosis as a differential diagnosis and perform chest CT and pulmonary function testing.

For airway stenosis, silicone stents (e.g. DUMON stent) and self-expanding metallic (SEM) stents (e.g. the Ultraflex stent) are widely used (5). Silicone stents have good expansion properties and are removable, so they have been the

first choice. However, silicone stents require rigid bronchoscopy and general anesthesia for placement. In Japan, few institutions use rigid bronchoscopy (6). In addition, migration may occur when silicone stents are placed in the trachea or bronchus (7). In contrast, SEM stents can be placed using video-bronchoscopic or radioscopic guidance under local anesthesia. Metallic stents have little risk of migration, but their expansion power is weak. In addition, SEM stents are less readily removed.

A new SEM hybrid stent, the AERO stent, was approved by the US Food and Drug Administration in 2007 and has been widely used in the US and other countries since then. However, in Japan, this stent was only launched in late 2016. The AERO stent combines the best features of silicone and metal stents with a completely covered nitinol framework. The potential advantages of this stent are (1) insertion using flexible bronchoscopy, (2) easy removal, and (3) strong expansion properties (8). There are two types of AERO stent. One uses an over-the-wire (OTW) delivery system and the other a direct visualization (DV) delivery system. In the OTW delivery system, the stent is introduced to the lesion by a guidewire through a rigid tube or a tracheal tube under fluoroscopic guidance. In contrast, with the DV delivery system, the stent is introduced to the lesion under bronchoscopic guidance.

In Case 1, we considered ACC as a differential diagnosis at the first examination. In such cases, we plan to remove the stent after radiotherapy; therefore, we considered the DUMON and AERO stents as candidates in this patient. Considering that the stenosis was in the middle part of the trachea, we chose the AERO stent because of its solidity.

In Case 2, we chose the AERO stent because the lesion was present in the upper part of the trachea and the stability of the stent was important at this site. We placed the AERO stent using radioscopic guidance under general anesthesia without rigid bronchoscopy because the patient had cervical spondylosis. However, we administrated VA ECMO preoperatively and were prepared for rigid bronchoscopy in case of emergency. At our institution, as a rule, we perform airway stenting using rigid bronchoscopy because we consider obstructive or hemorrhagic complications to be able to be more rapidly controlled with rigid bronchoscopy than with flexible bronchoscopy.

The most serious complications associated with airway stenting are infection, migration, and granulation tissue formation. The AERO stent reportedly carries an increased risk of infection compared with other stents, but it has the advantage of a decreased risk of migration (7). In our cases, we performed bronchial lavage using flexible bronchoscopy two to three times after stent placement, and infection and migration were not observed. In Case 1, at follow-up six months after placement, there were no observations of migration of the stent, bleeding, or infection. In Case 2, at one month after stent placement, there were no observations of stent migration or bleeding.

In Japan, the long-term safety of the AERO stent is unknown because this stent has only recently been released in this country and few cases have been reported. The accumulation of a meaningful number of cases is expected in future.

Conclusions

We reported our experience with two cases in which a new hybrid stent, the AERO stent, was effective for tracheal stenosis due to a malignant tumor. This is the first report in Japan that shows the efficacy of the AERO stent for stenosis. Tracheal stenosis is difficult to diagnose, and patients with refractory cough and dyspnea should be carefully examined and undergo CT or bronchoscopy.

The authors state that they have no Conflict of Interest (COI).

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