Salvage surgery of nonremovable metallic stent for adenoid cystic carcinoma and the subsequent difficulty of airway management

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Abstract
In 1999, a 60-year-old female diagnosed with tracheobronchial adenoid cystic carcinoma, was rejected due to the high risk reconstruction surgery of the carina. Our interventional radiological doctor implanted metallic stents for bilateral bronchial stenosis (right: Ultraflex, left: Spiral Z stent) to prevent endobronchial edematous asphyxia during radiation therapy. Radiotherapy showed that the tumor had decreased in size. The migrated right metallic stent was removed but the left one remained because it could not be removed. She had been unevenful for 3 years after the metallic stent implantation. In the 4th year, bronchoscopic balloon dilatation therapy had been performed for the produced dyspnea due to the exuberant granulation once every year for 4 years. The left lung had been destroyed by resuscitate pneumonia, thus we performed a pneumonectomy as salvage surgery using a cardiopulmonary bypass. The postpneumonectomy syndrome, dysphagia and dyspnea had confused her. Bronchoscopic balloon dilatation therapy had been continued for the right single airway stenosis every three months for 2 years. Eventually, she died of respiratory failure due to the recurrence of the disease after 12 years of treatment. We should require use of a silicone stent for the low grade malignancy of tracheobronchial airway stenosis. Eventual open thoracotomic removal should be done for management of the long-term complications of nonremovable metallic stents.

Key Words: salvage surgery, adenoid cystic carcinoma, metallic stent, airway stenosis

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Introduction
Early on, metallic stents were frequently used for tracheobronchial stenosis, but found to cause several long-term complications. Hence, silicone stents are now usually chosen as the first stent, while metallic stents are mainly used for the palliative care of patients with a short life expectancy. For an inoperative critical airway obstruction (bilateral bronchial stenosis) and/or end-stage cases with a malignancy, we succeeded in improving the patient’s severe dyspnea by the combination of bilateral bronchial stent dilatation, and this interventional therapy may be approved as an option for the temporary remission therapy of carina stenosis and good quality of life along with improving the severe dyspnea for 3 months until the patient’s death1.

On the other hand, in the case of bilateral tracheobronchial stenosis due to a low grade malignancy of an adenoid cystic carcinoma (ACC), we had implanted not only silicone, but also a metallic stent. We report the unusual events of long-term complications and the difficulty of airway management due to a 12-year-old implanted metallic stent. Eventually, salvage surgery, a pneumonectomy, was performed on the destroyed lung due to the fractured nonremovable metallic stent with cardiopulmonary bypass.

Case report
A 58-year-old woman with a cough, fever, and sputum was diagnosed with pneumonia based on a chest X-ray in January, 1999 (Fig. 1a). She was diagnosed with bilateral tracheobronchial adenoid cystic carcinoma (ACC) in our hospital in February, 1999 (Fig. 1b). The computed tomography scanning of the chest showed the bilateral tracheobronchial airway stenosis which the tumor occupied in the carina and left entire main bronchus (Fig. 2a), and the tumor extended to the left lower bronchus with extramural growth (Fig. 2b). We informed the patient of the high risk surgery involving tracheobronchial resection and carina reconstruction along with the left pneumonectomy because of the anatomical limitation of the long segmental disease of the left main bronchial tumor. She had rejected the aggressive surgery with a high mortality and morbidity and provided informed consent for nonsurgical therapy such as radiotherapy and bilateral bronchial interventional therapy for the unresected low grade carcinoma...
malignant tumor. We considered the conservative therapy and consulted a radiological doctor on this patient’s radiotherapy and interventional therapy, who selected a metallic stent insertion for the prevention of asphyxia due to endobronchial edema during radiotherapy. In those days, we could not expect that an expandable stent insertion was hardly indicated for this case, because the metallic stent would have been in a nonremovable status due to the growth of the granulation and endobronchial fibrosis. The exact reason why attending doctors inserted a non-covered expandable metallic stent was because of a long-segmental stenosis such as from the left main to the lower bronchus, and that the non-covered Spiral Z stent would less obstruct the orifice of the left upper bronchial bronchus. In our current concept, we should employ a soft silicone stent in consideration of its removal or select a covered metallic stent with an opened hole so not to cover the orifice of the left upper bronchus. However, even now, the appropriate stent selection might be hardly indicated for this case with a long segmental stenosis.

In February, 1999, under general anesthesia, snaring, laser ablation, and 99.5% ethanol injection were performed for the endotracheobronchial tumor, and the bilateral stenosis was released. Fig. 3 (a, b) shows debulked endobronchial findings before the stent insertion.

In March, 1999, under general anesthesia, with x-ray fluoroscopy, the stenotic segment of the left main bronchus was 5.7 cm in length, and after dilatation of the stenotic bronchus with a balloon catheter, a stainless Spiral Z stent (10 mm diameter, 57 mm length, Medico’s Hirata, Osaka, Japan) was inserted across the lesion starting from the distal part of the left lower bronchus and ending at the proximal main bronchial orifice of the carina (Fig. 3c, Fig. 4). The stenotic segment of the right main bronchus was 2.0 cm long, similarly, after the balloon-dilatation, an expandable metallic stent (12...
mm diameter, 20 mm length, Ultraflex, Noncovered Microvasive Stent [Boston Scientific, Natick, MA] was inserted in that position (Fig. 3d, Fig. 4). The location of the tumor and inserted stents are shown in Fig. 4.

X-ray fluoroscopy showed an expanded bilateral bronchi that alleviated the airway obstruction (Fig. 5a). After irradiation therapy (total of 40 Gy), the obstructive pneumonia had diminished (Fig. 5b) and complete remission of the tumor was observed (pathological negative findings resulted in endobronchial mucosa by bronchoscopic biopsy) (Fig. 6a, 6b). In July, 1999, she had anterior chest pain with a cough and dyspnea when she moved, the right migrated metallic stent was endoscopically removed (Fig. 6c), but the left metallic stent could not be removed because of its epithelialization and being covered with endobronchial fibrosis by irradiation (Fig. 6d). In December, 2002, she had a rest-time dyspnea and sever cough caused by the tracheobronchial stenosis due to migration of the left unremovable metallic stent and exuberant granulation tissue (Fig. 7a). We did not suggest removal of the stent-wire under a rigid bronchoscope because of the high risk of unexpected endobronchial bleeding from the pulmonary artery and aorta due to the stent-migration. In preparation of the setting up of the percutaneous cardiopulmonary support, high-frequency wave ablation of the granulation tissue and bronchoscopic balloon dilatation intervention therapy with a 7-Fr, 10-mm angioplastic balloon catheter (Meditech, Watertown, MA, USA) were performed under general anesthesia every year from June, 2003 to August, 2006. In November, 2006, the effectiveness of balloon dilatation had not been able to be maintained even for 3 months (Fig. 7b). We performed many biopsies from the left main bronchus during the balloon dilatation therapy, but there was a pathological finding of granulation tissue and no evidence of recurrence. In April, 2007, she had a fever, cough, and rest-time dyspnea in bed, because of the left destroyed lung after obstructive pneumonia of the
upper lobe. Three-dimensional computed tomographic (3D-CT) scanning of the chest revealed the migrated and nonremovable metallic stent in the left main bronchus (Fig. 8a), and a bronchoscopy showed the fractured stent-wire sticking into the right-sided proximal tracheobronchial wall, but the bronchoscopy could not evaluate the right-sided bronchial airway (Fig. 8b). In June 2007, a pneumonectomy as salvage surgery with removal of the metallic stent from the left main bronchus was planned. After a median full sternotomy and under cardiopulmonary bypass of cannulation into the left femoral arterial (14 Fr), right atrium (32 Fr), and SVC (28 Fr), although the left lung hilar site had changed to a bulky stone-like hardness by irradiation fibrosis, the distal left main bronchus was divided with scissors. The residual fractured stent wires were removed through the divided bronchial edge, which was closed with 3-0 prolene sutures. Endoscopically, we removed the proximal endobronchial stent wires by grasping the stent-wire with alligator for-
Fig. 6. a: Bronchoscopic view displaying an open tracheobronchial airway after bilateral metallic stent implantation and irradiation, however, the right Noncovered Microvasive Stent had migrated in the proximal direction (R).
b: Chest CT displaying the reduction of tumor and complete response.
c: The migrated noncovered Microvasive stent implanted in right main bronchus was bronchoscopically removed.
d: Bronchoscopic finding showing Spiral Z stent implanted in left main bronchus (L), which could not be bronchoscopically removed because of epithelialization and covered with endobronchial fibrosis by irradiation.

Fig. 7. a: Bronchoscopy displaying severe central stenosis of carina due to granulation formation.
b: Bronchoscopic view displaying the right main bronchial airway after balloon dilatation therapy. The proximally-migrated metallic stent did not allow the bronchofiberscope to enter the left main bronchus.
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Fig. 8. a: 3D-CT scanning showing the left metallic stent had proximally migrated to the right tracheal wall and clogged the right main bronchial orifice. 
b: The bronchoscope could not enter the distal tracheobronchial tree and showed the wires of the migrated stent at the proximal carina level.

Fig. 9. a: 3D-CT scanning displaying the nonremovable, unburied, residual stent. There was no stent-wires in the airway after intraoperative bronchoscopical removal. 
b: The right tracheobronchial airway was open and there was no obstacle due to stent-wires in the tracheobronchial tree after salvage pneumonectomy.

Fig. 10. The resected left lung. Box numbers 1, 3, and 4 show a recurrence of the adenoid cystic carcinoma and lymph nodal metastasis.
a bronchial intervention every three months for about 2 years, however, the patient’s quality of life could not be dramatically improved. In February, 2010, the patient was dead of respiratory failure due to the recurrence of the disease after about 12 years from the first diagnosis of ACC.

Discussion

Adenoid cystic carcinoma (ACC), developing in the trachea/bronchi, is a relatively rare low-malignancy cancer of bronchial gland origin. Resection of the tracheal carina and reconstruction with a double-barreled carina-plasty and the number of reports on surgically resected cases of ACC has been increasing[3]. A characteristic of the ACC arising from the trachea is that even when the trachea is resected with upper and lower margins of 1 cm from the macroscopic or palpable tumor, the surgical stump is sometimes tumor cell-positive[4]. In such cases, postoperative radiotherapy seems indispensable because of its high radiosensitivity of the ACC[3].

This case required a surgical tracheobronchial carina resection and tracheobronchoplasty, and also with or without pneumonectomy because of the anatomical limitation of the long segment of the left main bronchial tumor, even though it might mean an almost unresectable case and operative procedures requiring a high skill. Although we informed the patient of the high risk of the tracheobronchoplasty and a possible unresectable surgery due to the anatomic limitation, the patient rejected the aggressive surgery and selected the non-surgical therapy of radiation and implantation of a tracheobronchial stent, together with laser ablation and bronchoscopic balloon dilatation.

Currently, a silicone stent should be selected when considering removal before radiotherapy. However, in those days, as there was no determined indication of appropriate stent selection for a carina-type tumor with bilateral bronchial stenosis, the expandable metallic stent implantation for bilateral bronchial stenosis was performed. Especially, the carina-stenotic tumor was thought to be a difficult case compared to the one-sided segmental stenosis of the tracheobronchial airway. It was a fact that whether or not the Y-type silicone stent might exactly fit for the bilateral bronchial stenosis, some difficulties of preserving the structural strength regarding the inside diameter and the longitudinal straightness might occur. A metallic stent had been selected that was the combination of two different types of stents, i.e., the metallic Ultraflex stent for the right stenosis and the spiral Z-stent for the left one. We did not consider the long-term complication in this case due to the nonremovable metallic stent. On the other hand, even if a removable silicone-type stent had been used for the left bronchus, sooner or later, the problem of migration, kinking, and reduced structural strength compared to the metallic stent might have occurred. Needless to say, we should not have implanted the metallic stent for benign disorders, and also for a low grade malignancy such as ACC and in a remission case of definitive chemoradiation therapy for a malignancy. In this case, we used a metallic stent for the ACC before irradiation therapy to prevent postradiated edematous asphyxia, which resulted in the nonremovable status due to epithelialization and radiation fibrosis. Using metallic tracheal stents as a bridge to other therapies is not recommended.

Improvements in the stent design and the techniques of interventional bronchoscopy in recent years have extended the use of endoscopically deployed expandable metallic stents for patients with benign and malignant
airway stenosis. The advantage of these stents are ease of placement, good internal-to-external diameter ratio, and low incidence of stent migration. However, the disadvantages of metallic airway stents include difficult extraction, tendency for stent fatigue and fracture, and ingrowth of stent with a tumor or granulation tissue. A self-expandable metallic stent fracture is not uncommon in patients with tracheobronchial disease, and the fracture incidence dramatically increases 2 years after implantation, thus the metallic stent should be restricted to a highly selected population rather than all patients with a benign disease to specifically avoid situations in which long-term use of the stent is anticipated.

Complications due to metallic airway stents include granulation tissue formation, fracture of the struts, migration, and mucous plugging. When these complications result in airway injury or obstruction, it may become necessary to remove the stent. If there had been encroachment upon any nearby blood vessels, the pulmonary artery for example, any reckless attempt to remove or relocate the stent may result in a fatal outcome. For the long-term complication of a destroyed lung due to the nonremovable implanted metallic stent, which could not be removed by bronchoscopical traction with alligator forceps because of the fibrosis, an eventual pneumonectomy as salvage surgery, and open thoracic removal surgery should be done along with a cardiopulmonary bypass.

In 2005, an FDA Public Health Notification was disclosed about “Complications from Metallic Tracheal Stents in Patients with Benign Airway Disorders.” We have summarized this notification for dealing with a metallic stent-implantation. This notification focuses on patients with benign airway disorders, because the use of metallic stents in this patient population may preclude them from receiving future alternative therapies, such as tracheal surgical procedures or placement of silicone stents, after the metallic stent is removed. This patient population has a greater risk of serious complications than those with malignant disorders since the metallic tracheal stent is left in place longer. Removal of metallic stents can also result in serious complications, including mucosal tears, severe bleeding, re-obstruction, respiratory failure with the need for postoperative mechanical ventilation, and tension pneumothorax. If the stent is removed in pieces due to device failure or fracture during removal, this can lead to unwanted permanent incorporation of retained stent fragments in the tissue. If a metallic tracheal stent is the only option for a patient, insertion should be done by a physician trained or experienced in metallic tracheal stent procedures. If removal is necessary, the procedure should be performed by a physician trained or experienced in removing metallic tracheal stents. In this case, unfortunately, the nonremovable fractured metallic stent resulted in the eventual formation of a destroyed lung, thus salvage surgery for removal and infection control should be safely performed along with a cardiopulmonary bypass.

When carefully considering the appropriate indications for use and selecting patients, and also after stent insertion, we have to have a better follow up and proper management of the implanted stents. If we cannot perform the eventual removal surgery, we should not insert the metallic stent. When we have to use a metallic stent for a tracheobronchial stenosis, only do it for a malignancy with a 3-month prognosis, not for a benign disease, low grade malignancy or pre-state radiation therapy.

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