Brief Report

Broncho-vascular fistulas from self-expanding metallic stents: A retrospective case review

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Abstract:

To highlight a potentially fatal complication of broncho-vascular fistula arising from the self expanding metallic stent (SEMS) placement. We retrospectively analyzed five patients with benign and malignant airway diseases, who developed tracheo/broncho-vascular fistulas following SEMS placement in our tertiary care setting. All patients received either Wallstent or Ultraflex® stent (Boston Scientific, Natick, MA) between 1999 and 2007. All patients had received adjunct therapy such as balloon bronchoplasty, laser therapy or electrocautery. Most patients presented with massive hemoptysis. A total of 483 SEMS were placed during this period. SEMS placement can be complicated by Broncho-vascular fistula formation. True incidence and precise time interval between the insertion of stent and onset of this complication is unknown. Additional therapeutic modalities to maintain stent patency may enhance the risk of fistula formation. SEMS should only be used in a select sub-group of patients, after exhaustive evaluation of other treatment options. These cases provide evidence that broncho-vascular fistulas can develop at any time following SEMS placement, suggesting the need for a more cautious approach, especially while using them for a long term management. In benign airway disease, the stent should be removed as soon as healing has taken place.

Key words:

Benign airways disease, bronchial stents, complications, endobronchial growth, ultraflex, wall stent

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Management of central airway obstruction, from both benign and malignant diseases, is one of the most challenging aspects of respiratory medicine. Airways stenting is a reliable palliative approach to maintain airway patency. Endobronchial stenting of airways has made long strides since its beginning in 1960s. Given the ease of placement, self-expanding metallic stent (SEMS) had become very popular in recent past. Unfortunately, the initial enthusiasm in SEMS placement was dampened by a frequent complication of excessive granulation tissue formation and difficulty in its removal.[1] Incidentally formation of broncho-vascular fistulas (BVF) is also a major complication of SEMS yet reported less frequently. In fact, Food and Drug Association (FDA) published a warning in favor of a more restrained approach for airway stent placement.[2] We describe a series of five cases treated with SEMS placement and suffered with BVF. Purpose of our report is to highlight the complication by supporting our experience with the review of the literature.

Case Reports

Case 1

A 50-year-old male, underwent double lung transplant for idiopathic pulmonary fibrosis (IPF). Two weeks following his transplant, he was readmitted with acute onset of severe shortness of breath. His Computed tomography (CT) of the chest revealed a small air collection adjacent to right anastomotic site with bilateral pleural effusions. His echocardiogram demonstrated large circumferential pericardial effusion with tamponade physiology. The pericardial fluid was evacuated by a pericardial window and a right sided chest tube.

Flexible bronchoscopy (FB) was performed the same day, which confirmed bilateral bronchial dehiscence, worse on the right, with a 1.5 cm defect of antero-lateral wall of right main stem bronchus with separation of suture line from donor bronchus [Figure 1]. There was a free moving flap and full thickness necrosis of the wall which extended down to bronchus intermedius. A 14 mm × 40 mm uncovered Ultraflex® SEMS was deployed over a guidewire in the right main bronchus to bridge the dehiscence. With bronchoscopic forceps, the stent was gently repositioned firmly against the secondary carina in an effort to cover the defect. A 12 mm controlled radial expansion (CRE) balloon was used to dilate the stent across the area of the anastomosis. The balloon of smaller diameter was used to avoid undue high atmospheric pressures at the site of disruption. At the end of the procedure, the stent was in a visibly good position.

Patient was doing well at home for 3 weeks when he experienced a sudden massive hemoptysis. In emergency, his airway was noted to be filled with blood and he had right sided hydropneumothorax. He died shortly after arrival in the emergency department. Considering his recent intervention and stent placement, we concurred that BVF was the most likely mechanism of his massive hemoptysis. Unfortunately, family refused an autopsy.

Case 2

A 73-year-old female, ex-smoker with 4 pack-year of smoking history was diagnosed with ovarian adenocarcinoma at 67 years of age. She underwent surgery for local spread of tumor in addition to chemotherapy. Five years following the initial diagnosis she developed a lung lesion and supraclavicular lymphadenopathy. Biopsy from both lesions revealed adenocarcinoma. She received chemotherapy along with external beam radiation. She presented with recalcitrant cough and left lower lobe collapse due to stenosis of the left main stem bronchus from extrinsic compression. A covered 12 mm × 40 mm Ultraflex® SEMS was placed in the left main stem endobronchial lesion with symptomatic and radiographic improvement. Four months following the procedure, the patients developed recurrence of her cough; FB revealed ingrowths of granulation tissue involving the distant third of the stent and an ulcer in lower third of trachea. Bronchial brush cytology was negative for malignant cells. She underwent two separate endobronchial multimodality treatment for debulking of the granulation tissue using electrosurgery, cryoablation, balloon bronchoplasty, and intrabronchial steroid injection. She started experiencing hemoptysis 4 months after her last procedure and a year after initial stent placement. Bronchoscopy revealed blood oozing from stent site in left main stem bronchus. Her CT of chest with contrast showed loss of the tissue plane between the stent and the left pulmonary artery with metallic stent eroding into the left pulmonary artery [Figures 2 and 3]. She continued to have recurrent hemoptysis for next 1 year and eventually passed away from exsanguination.

Case 3

A 49-year-old man underwent double lung transplant for Idiopathic Pulmonary Fibrosis and pulmonary hypertension. 10 months following the transplantation he developed breathlessness and wheezing. Bronchoscopy revealed a near total obstruction of the right main bronchus by a whitish, exophytic lesion, which was excised and airway was dilated. Biopsy of the endobronchial lesion revealed an aspergilloma. The patient was appropriately treated with systemic and topical antifungals. Subsequently, he underwent Ultraflex® SEMS placement and brachytherapy (dose - 7.1 Gy × 2) in the right bronchus for excess granulation tissue. He improved initially but two months later developed worsening hemoptysis. While hospitalized, he developed cardiac arrest following massive hemoptysis and required an urgent right pneumonectomy. Decision to perform pneumonectomy was based on clinical suspicion of BVF, a diagnosis compatible with his presentation. Intraoperatively, 1 cm defect in posterior right pulmonary artery was noted, communicating with the SEMS in the right bronchus intermedius. Stent could be grasped from the pulmonary artery but unfortunately he expired the next day.

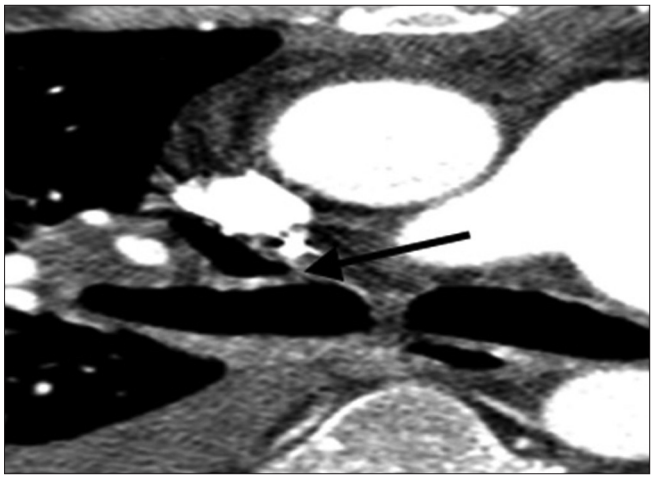

Figure 1: Anastomotic dehiscence – Right main stem bronchus (arrows)

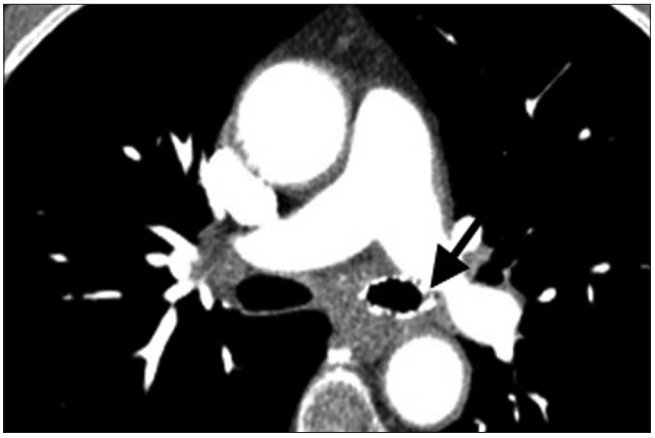

Figure 2: SEMS abutting the left pulmonary artery

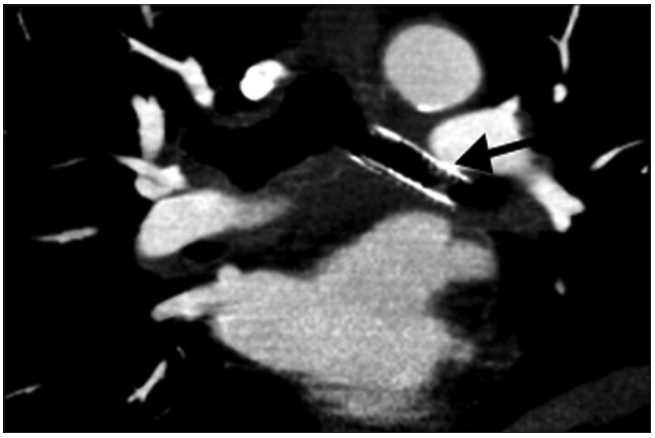

Figure 3: Coronal view showing loss of tissue plane between SEMS and pulmonary artery

Case 4

A 53-year-old male underwent right lung transplantation for severe chronic obstructive pulmonary disease (COPD). He subsequently developed right anastomotic dehiscence requiring Ultraflex® SEMS stent 14 mm × 40 mm placement in right main bronchus. Patient continued to have recurrent in-stent restenosis treated with Neodymium-doped yttrium

aluminum garnet (Nd-YAG) laser, balloon bronchoplasty and electrocautery between 14 and 39 months post stenting. Two months following his last intervention, he presented with wheezing. Electrocautery was attempted to dilate his anastomotic stenosis, but was complicated by 400 ml of bleeding. On review, his CT scan of chest demonstrated metallic stent, bridging the anastomosis closely applied to posterior aspect of right main pulmonary artery [Figure 4]. He was eventually discharged home and died within a few days from massive hemoptysis.

Case 5

A 65-year-old female, diagnosed with non-small cell lung cancer developed respiratory distress and hypoxia requiring a bronchoscopy. It revealed tracheal narrowing due to extrinsic compression from the tumor, in addition to complete occlusion of the left main bronchus due to endobronchial invasion. Extensive tumor debulking was performed using Nd-YAG laser and balloon bronchoplasty. A covered Ultraflex® SEMS 18 mm × 60 mm was placed into distal trachea extending into the left main bronchus. Patient completed her radiotherapy and chemotherapy course. Seven months post intervention patient, she started complaining of increasing wheeze and halitosis. Repeat bronchoscopy revealed tumor ingrowths into the stent, broken stent wires, and a tracheo-mediastinal fistula noted at distal end of the stent. Laser photoresection was done to maintain stent patency. A week later she developed hemoptysis and CT of the chest revealed pneumo-mediastinum from tracheo-mediastinal fistula with irregular medial contour of the aortic arch consistent with invasion of the aorta [Figure 5] requiring transition to hospice care.

Discussion

Between 1999 and 2007, total 483 SEMS were inserted at our institution. Five of these patients developed BVF and eventually succumbed to the complication. Average age of patients was 58 (49-73) years, male to female ratio of 3:2. Three patients had airway dehiscence following lung transplantation and two had malignant disease. All patients except one received additional endobronchial therapy including photocoagulation, cryotherapy, balloon bronchoplasty, or electrocautery to mange complications of the stent. Mean duration of fistula formation was 16 (1.5-65) months [Table 1].

Surgery is the best option for both benign and malignant central airway obstruction. However, often patients are not surgical candidates for a variety of reasons. Even after surgery number of patients with benign disease may develop either recurrence or surgical complications.[3]

Advent of SEMS with improved design and deployment has led to their increased used in a variety of tracheobronchial disorders.[4] SEMS are made from a nitinol base alloy and their advantages include, ease of insertion, shape memory to conform to tortuous airways, greater cross-sectional area and preservation of mucociliary clearance.[5] However, these advantages are often offset by numerous potential problems. Several studies have shown complication rates approaching 50%.[4,6] Common complications are mucus build up (38%) leading to infectious tracheobronchitis, halitosis, obstructing granulation tissues or tumor ingrowths (36%), and stent migration (12%).[7] Other complications reported are stent fatigue or fracture, complications associated with attempted stent removal,[8] trachea/broncho-esophageal fistula and BVF formation.[9,10] Incidentally, anastomotic dehiscence following lung transplantation is often treated with placement of SEMS. If left on place for long durations, these patients may develop stent obstruction from granuloma formation requiring further interventions.

True incidence of BVF formation is unknown as many patients experience massive hemoptysis at home and may not reach the hospital. There are at least 11 reported cases of tracheo/BVF with first generation SEMS, notably Gianturco stent.[11-13] We also noted four cases of similar complication with Palmaz stent in the pediatric population.[11,14] Fortunately, this complication

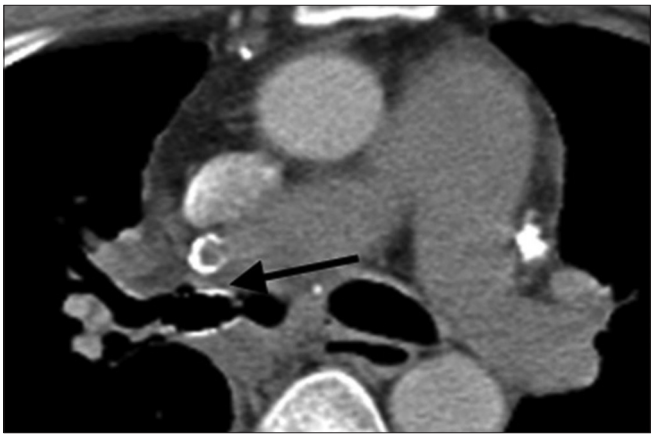

Figure 4: SEMS in right bronchus abutting right pulmonary artery

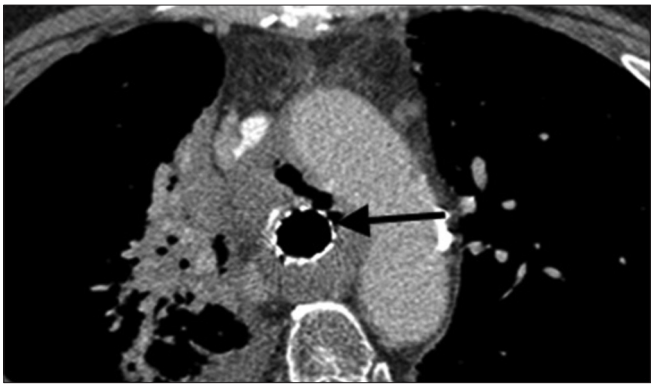

Figure 5: Tracheo-mediastinal fistula seen between SEMS and aorta (arrows)

Table 1: Diagnosis, stent types, and additional treatment

Age (Year)	Diagnosis	Stent	Additional therapies	Location	Duration (Months)	Alive (Y/N)
50	Dehiscence	U	None	RMS	1.5	N
73	Ovarian Ca	U	ES, C, BB, S	LMS	10	N
49	Stenosis	U	BB, ES, EBT	RMS	6	N
53	Dehiscence	U	ES, LPC, S	RMS	65	N
65	NSCLC	U	LPC, APC	Trachea	7	N

BB = Balloon bronchoplasty, ES = Electrocautery, APC = Argon plasma coagulation, S = Steroid injection, C = Cryotherapy, LPC = Laser photocoagulation U = Ultraflex stent, RMS = Right main stem, LMS = Left main stem

is uncommon with second generation SEMS. Katayama *et al*., reported a case of aorto-bronchial fistula with self-expanding nitinol stent.[15] Similarly, Shivo *et al*., described a case of fatal fistula between the trachea and brachiocephalic artery after insertion of covered Ultraflex® stent.[11]

This complication is reported anytime post-stenting, from early days or weeks to years after its placement. Our timescale is consistent with what described in literature. Interestingly, all the reported cases in literature developed fistula after stent placement in either Trachea or left main bronchus. In contrast, three patients in our series had right main stem stent placement, one each in trachea and left main bronchus.

There is no universally agreed practice regarding when an airway stent should be removed, particularly in benign airway disease.[16] There are conflicting evidences in literature regarding outcome after early stent removal.[10,17] None of the stents were removed in our group for either therapeutic reasons or because patients tolerated it initially.

Madden *et al.,* retrospectively reviewed complications of Ultraflex® stent in their patients between 1997 and 2005.[4] None of them had any Tracheo/BVF. Similarly no such complication was described by Gaissert *et al*.,[9] or Swanson *et al*.,[8] in their analysis of complications with metal stents. This difference can be due to the patient population, use of adjunct therapy, difference in stent sizing to cover the entire lesion and the degree of dilatation required for full deployment. Three of our patients were lung transplant recipients and most likely developed fistula at the site of airway and anastomosis. It is unlikely that anti-rejection medications played any role in formation of this complication as none of our patients were on Rapamycine. Incidentally, late dehiscence related to use of Rapamycine has been reported in the literature.[18] One of our patient received endobronchial brachytherapy receiving total 14.2 Gy dose. Besides, all but one of our patients underwent extensive adjuvant treatment. These therapies certainly played a role in weakening vascular and airway walls. Cordasco *et al*., made similar observation suggesting prior laser or radiation therapy sufficiently weakens airway walls.[19]

Three of our patients underwent lung transplant with relatively better median survival and two had malignant endobronchial stenosis where stents were placed for palliative purpose. In lung transplant patients, stent placements were done with definite intention to treat. Although SEMS plays a useful role in malignant airway diseases, its role is less clear defined in benign endobronchial conditions. Nevertheless, self expanding metallic stents are increasingly employed in benign diseases, particularly where surgery is not an option. Gotway *et al*. noted significant improvement in airflow and spirometry after stent placement in benign tracheobronchial stenosis. Subsequent stent placements may provide additional improvements in pulmonary function.[20]

Conclusion

Although rare, SEMS placement can result in fatal complication such as BVF formation at any time after SEMS implantation. Despite its usefulness, these devices are best considered as last resort for tracheobronchial diseases. SEMS should only be used after ruling out other potentially safer options, especially for long term treatment in benign airways diseases especially among lung transplant recipients.

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