



Case report

Long-term tolerance of a fractured self-expanding metal stent in a patient with adenoid cystic carcinoma

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ABSTRACT

Self-expanding metals stent are used for recanalization of malignant CAO. Fracture of such a stent has been described. This prompts its removal. However removal of a metal stent is difficult and can be hazardous. Due to this US FDA advisory was released against their insertion in benign diseases where long life expectancy can expose the patients to the complications of prolonged indwelling time of these stents. We describe a case of adenoid cystic carcinoma who required SEMS placement for tracheal obstruction. However, stent fracture developed soon after its insertion causing patient to cough out stent pieces multiple times. In view of the challenge associated with removal of metal stent fixed to the tracheal walls at its proximal and distal ends, the stent was left in-situ. Patient has tolerated the fractured stent for 1-year and remains on follow-up without any complication or adverse event. This illustrates the long-term tolerance and safety of a fractured stent and helps to allay anxiety associated with this complication.

1. Introduction

Adenoid cystic carcinoma (ACC) often presents with central airway obstruction (CAO) whereby recanalization of the airway becomes the first priority. Once re-canalized, it's a low grade cancer with 5-year survival of 89% [1]. Although contraindicated in benign CAO due to difficulty in, or complications associated with removal, Self-expanding metal stent (SEMS) have been widely used for recanalization of malignant airway obstruction [2]. Successful placement prevents premature death from suffocation, and allows time for definitive cancer therapy to take effect [2]. Such stents have been reported to be safely tolerated in the airways for the median duration of 3.5 years [3].

Compared to silicon stent, SEMS offers the advantages of lower migration rate, better conformation to irregular airways, and greater ease of placement as it can be placed even via flexible bronchoscopy under moderate sedation [2]. However, its use is associated with complications such as retained secretions, bacterial colonisation of stent, granulation tissue formation, difficult retrieval, high risk of hazardous complication upon attempting removal, and stent fracture [2,4].

We report a rare case of 1 year tolerance of fractured stent in a patient with adenoid cystic carcinoma (ACC) of the trachea. In authors' knowledge, this is the first report of long-term tolerance of a fractured

SEMS.

2. Case report

A 66-year-old female presented with a 1-month history of gradually worsening shortness of breath and productive cough. She was a non-smoker with history of gastroesophageal reflux disease, obstructive sleep apnoea, and transient ischemic attack.

Her physical examination revealed inspiratory and expiratory wheeze. Chest X ray was normal. In view of persistent shortness of breath and normal radiograph, computed tomography pulmonary angiogram (CTPA) was done to rule out pulmonary embolism. CT scan showed an infiltrative mass measuring 3.0 x 2.5 x 2.5 cm in the right lateral wall of the lower trachea about 1 cm above the carina causing significant airway obstruction (Fig. 1a). The patient underwent rigid bronchoscopy with laser resection and tracheal stenting under general anaesthesia. After debulking, a 20 x 60 mm Ultraflex covered SEMS was placed in the trachea.

Histology revealed adenoid cystic carcinoma. Subsequent staging did not reveal extra-tracheal spread. She was offered tracheal resection and anastomosis. However she declined due to risks associated with the surgery. She also rejected concurrent chemo radiation, and opted only

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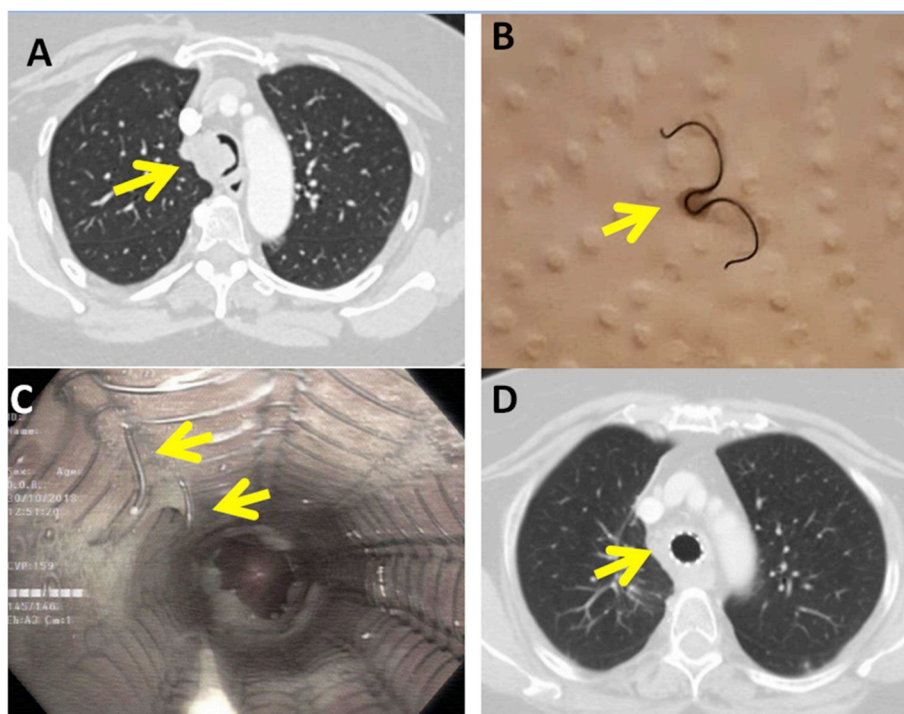


Fig. 1. 1A) CT scan showing adenoid cystic carcinoma with near complete tracheal obstruction. 1B) Pieces of metal stent (nitinol wires) coughed out by the patient. 1C) Bronchoscopy showing the fractured wires (arrows) of the metal stent in the trachea. 1D) Follow up CT scan showing the fractured stent in the trachea with maintained tracheal patency.

for radiotherapy, which was planned for 66 Gy in 33 fractions.

One day prior to radiotherapy, she coughed out 2 small stent pieces mixed with sputum (Fig. 1b). Bronchoscopy showed breaks in the wires of the stent in the left lateral aspect at 3 places (Fig. 1c). The stent was otherwise well placed with good scaffolding effect on the trachea, maintaining tracheal patency. Decision was made to leave the stent in place in view of the maintained tracheal patency, risk of airway compromise from tumour if the stent was removed, and anticipated difficulties with stent removal due to granulation tissues at both the proximal and distal ends of the stent.

She was given cough suppressant and radiotherapy was started. She had another episode of coughing out stent pieces 3 months after stent insertion. She was offered rigid bronchoscopy and removal of the stent but declined. Inspection flexible bronchoscopy showed good position of stent with stable areas of stent fracture. CT scan also revealed a patent tracheal stent with no focal soft tissue lesion seen within the trachea (Fig. 1d). She has remained well with the fractured stent in-situ for 1 year at the time of this writing.

3. Discussion

This case illustrates the long-term tolerance of a fractured SEMS in a patient with ACC that was managed with laser resection and SEMS placement, followed by radiation therapy. SEMS are only advocated for malignant CAO as the short life expectancy is expected to obviate the need for removal, and exposure to stent related complications. However, life expectancy of patients with ACC is long. Ideally, patients with ACC should be evaluated for curative tracheal resection and anastomosis prior to placement of SEMS, as SEMS can become embedded in the airway mucosa with time and may negatively impact surgery [5]. While surgical resection is the treatment of choice for ACC, some patients are inoperable, unfit, or prefer not to undergo surgery. Placement of airway stent (SEMS or silicone) is the only option for such patients when presenting with CAO [5]. In this case, diagnosis of ACC had not been established at the time of recanalization of trachea and placing a silicone

stent was deemed challenging, hence SEMS was inserted.

Development of stent fracture led to the anxiety and feelings of regret among the managing physicians. When the patient developed stent fracture, not only was there physical breakage of the stent, she also had expulsion of nitinol stent fragments, which has been described rarely [6]. A stent fracture rate of 9.5% was reported by Chung et al. in their analysis of 211 patients who underwent SEMS placement [7]. Most patients with SEMS fracture present with an exacerbation of dyspnoea or cough, as seen in our patient [8]. SEMS fractures were also found to be more common in patients with benign airway obstruction. The reason postulated was the shorter life expectancy of patients with malignancy may not be long enough for this complication to develop [8].

Proposed causes of stent fractures include repetitive coughing, oesophageal compression during swallowing, metal fatigue, granulation, and shearing forces [6,9, and 10]. According to Chung et al., in 2008, an independent predictor for SEMS fractures was tortuous airway. The inability to maintain the ideal architecture and alignment, with increased shearing force and subsequent granulation tissue formation, might lead to fracture [8]. In our case, the patient did not have a tortuous airway. Although the repetitive coughing likely played a contributory role, whether this was related to a manufacturing defect remains unknown. It was also unusual that she developed stent fracture so soon after implantation (22 days), as most SEMS fractures were detected 500–1000 days post implantation, with increasing incidence of stent fractures with time [8].

The description of stent fracture management is sparse. SEMS fractures have been managed with removal of the fractured stent, implantation of another stent or observation [9]. Removal of SEMS via bronchoscopy can be challenging due to embedment of metal into the tracheal mucosa. Both rigid and flexible bronchoscopic techniques have been used to deal with this complication. Removal can either be in a piecemeal fashion or as a whole [2, 11, and 12]. However, the removal of fractured stent is not without risks. These include fragment retention, mucosal tearing and haemorrhage, repeat obstruction, pneumothorax, and the need for mechanical ventilation [2, 12].

Table 1
Serial lung function tests.

Date	Ref	17/05/2019		17/05/2019		19/07/2019		20/08/2019		9/10/2019	
		Pre Measured	Pre % Ref	Pre % Ref	Pre % Ref	Pre % Ref	Pre % Ref	Pre % Ref			
FVC	Litres	2.50	2.10	84	78	79	74				
FEV1	Litres	2.04	1.64	80	84	74	64				
FEV1/FVC	%	82		78	88	76	86				
FEF25-75%	L/sec	3.81	1.64	43	65	36	29				
PEF	L/sec	4.58	3.65	80	96	53	41				

The removal should be planned based on the difficulty level judged by the stent and airway interphase. A free stent in the airway or a 'floating stent', is easy to remove by grasping it with rigid forceps followed by rolling and removing it. However, the partially covered stents as in our case are difficult to remove after 2 weeks due to significant in growth of granulation tissue (GT). These stents should only be approached with multidisciplinary planning, including radiologic and bronchoscopic evaluation, in concert with cardiothoracic surgery and extracorporeal circulation back up. GT should be treated with cryotherapy or argon plasma coagulation. This reduces the stent incorporation into the airway wall, making them easier to remove. The stent should be removed in a piecemeal fashion, with short segments removed at a time. Bleeding usually occurs and can lead to a breach in the airway or vasculature requiring emergent thoracic surgery. If significant bleeding or critical airway obstruction occurs during the removal, patient should be placed on extracorporeal circulation and undergo thoracic surgery to remove the remaining pieces of the stent by directly opening the trachea [13]. Hence, in some cases, keeping SEMS in-situ may be a safer option.

Assessment of the stability of fractured stent has not been previously described. Based on serial CT imaging (Fig. 1d) and inspection bronchoscopy, our patient's fractured stent has remained patent and stable without causing any symptoms to patient. Lung function has also remained preserved (Table 1).

In conclusion, our case illustrates the long-term tolerance and safety of a fractured stent for a year and helps to allay anxiety associated with this complication. This conservative approach may avert complications in frail patients with multiple co-morbidities who may otherwise be subjected to bronchoscopic stent removal.

Declaration of competing interest

S.H., S.K.G., A.W.K.N., D.Y.H.T., A.L.Y.H., A.C.K., N.N.S.Z, J.A. and A.V have no competing financial interests to disclose.

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