Silicone Tracheobronchial Stent: A Rare Cause for Bronchoesophageal Fistula and Distortion of Airway Anatomy

Abstract
Silicone tracheobronchial stents are being increasingly used in a large number of patients for the treatment of tracheal stenosis. One very rare complication due to tracheobronchial stenting is bronchoesophageal fistula (BEF), which has been associated with the use of metallic stents. We report intraoperative management of a patient undergoing repair of a BEF following previous insertion of a silicone Y-stent that is soft in texture and has not been implicated for this complication till date. In addition, misalignment of this silicone tracheobronchial Y-stent resulted in a tracheal mucosal bulge proximal to the stent that vanished after its removal.

Keywords: Airway distortion, silicone stent, tracheobronchial fistula

Introduction
Tracheal stenosis is common sequelae of prolonged endotracheal intubation or tracheostomy.[1] A large number of these patients are being treated with tracheobronchial stents due to improved design and advanced technology to aid in their insertion. Numerous complications such as partial or complete airway obstruction, stent migration, mucostasis, and resulting infections have been reported in the literature.[2-3] Formation of fistula is a rare complication that has been associated with the use of metallic stents.[4] Silicone stents have not been implicated for this complication till date. This case report describes intraoperative management of a patient undergoing repair of a bronchoesophageal fistula (BEF) following the previous insertion of a silicone Y-stent and distortion of the airway anatomy because of the presence of stent. A written informed consent to publish this case report was obtained from the patient.

Case Report
A 26-year-old, 45 kg, 172 cm tall male with 6 months history of cough and expectoration of food particles following oral ingestion of food was admitted for surgical repair of BEF that developed due to in situ silicone tracheobronchial Y-stent inserted for the management of post tracheostomy tracheal stenosis. As per the records, the inserted tracheobronchial Y-stent had tracheal end diameter of 18 mm and bronchial end diameter of 14 mm.

The patient complained of occasional bouts of dry cough while sitting, which aggravated in the supine position. Preoperative examination showed normal vital signs and oxygen saturation of 95% on room air. Chest X-ray revealed patchy nodular opacities in the mid and lower zones of the left lung suggestive of chronic aspiration pneumonitis and presence of a tracheobronchial Y-stent. Contrast-enhanced computerized tomography (CT) confirmed a narrow communication in subcarinal region between proximal left main bronchus and esophagus abutting Y-stent posteriorly. A posterior mucosal bulge in the trachea was also noticed at the proximal tracheal end of the stent with marginal luminal narrowing [Figure 1a and b].

The patient’s tracheal dilatation and silicone tracheal stent placement for tracheal stenosis were done 5 years back. This was replaced with silicone tracheobronchial Y-stent following distal migration of tracheal stent. The tracheobronchial Y-stent was removed after 1 year; however, it was required to reinsert the following reappearance of symptom. Since then, he was asymptomatic on follow-up until last 6 months.

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How to cite this article: Kumar B, Munirathinam GK, Puri GD, Mishra AK, Arya VK. Silicone tracheobronchial stent: A rare cause for bronchoesophageal fistula and distortion of airway anatomy. Ann Card Anaesth 2017;20:355-8.
The key perioperative airway management issues for this case were (1) prevention of contamination of tracheobronchial tree from gastric content till isolation is achieved. (2) Selection of appropriate endotracheal tube (ETT) and its negotiation through the tracheobronchial stent. (3) Risk of increased leakage or aspiration through BEF in case silicone tracheobronchial Y-stent needs removal to facilitate passage of left-sided double lumen tube (DLT).

The surgical plan for this patient was to do BEF repair through left posterolateral thoracotomy and to do feeding jejunostomy. After consultation with surgeon and pulmonologist, our 1st plan was to place a 6 mm ETT endotracheally above the stent followed by advancing it through the stent into the right main bronchus under fiberoptic control. The standby plan was to remove the stent and use right-sided DLT under fibreoptic bronchoscopy (FOB) guidance. In worst possible situation of inability to ventilate or maintain saturation, a femorofemoral cardiopulmonary bypass (CPB) was kept ready as standby to start within few minutes. The patient was kept fasting for 8 h and received aspiration prophylaxis with oral ranitidine 150 mg and metoclopramide 10-mg preoperatively. On examination, no endotracheal intubation difficulty was anticipated. Airway management cart for this case included various sizes of ETTs, right-sided DLTs, flexible, and rigid bronchoscope. After instituting electrocardiogram and pulse oximetry, a large-bore intravenous access and invasive arterial blood pressure monitoring were established under local anesthesia inside the operating room. A 16-G nasogastric tube was inserted into the stomach in anticipation of the stomach distention through BEF following positive pressure ventilation. Anesthesia was induced with fentanyl 2 mcg/kg and titrated doses of propofol following preoxygenation with 100% oxygen. After confirming mask ventilation and absence of tidal volume loss into the stomach, suxamethonium 1.5 mg/kg was administered to facilitate endotracheal intubation under direct laryngoscopy using a 6 mm Portex ETT (Smith medical Australasia Pvt. Ltd., Brisbane QLD 4113, Australia). This ETT could not be advanced beyond 20 cm mark; hence, it was withdrawn by about 2 cm, and a FOB was inserted through it to visualize possible cause of resistance encountered. The FOB showed proximal anterior border of the stent and posterior tracheal mucosal bulge, obscuring the visualization of posterior margin of the stent [Figure 2]. An attempt to negotiate FOB across this mucosal bulge into the stent lumen failed. Considering malposition of the stent, a decision to remove it was taken. Hence, ETT was removed, and a 12 mm rigid bronchoscope was inserted into the trachea while maintaining spontaneous respiration and adequate depth of anesthesia. Rigid bronchoscope again confirmed posterior tracheal mucosal bulge proximal to the stent [Figure 3a]. The stent was removed using forceps through the rigid bronchoscope. During rigid bronchoscopy, ventilation was maintained manually with low tidal volume, and 100% oxygen using ventilation port of the rigid bronchoscope and peripheral oxygen saturation was maintained above 95%. Anesthesia was maintained using continuous intravenous infusion of propofol. The posterior tracheal mucosal bulge disappeared following stent removal [Figure 3b]. Subsequently, trachea was intubated with a 37F right-sided DLT under FOB.
guidance. The proximal margin of bronchial cuff entering into the right main bronchus and alignment of the right upper lobe ventilation slot against right upper lobe bronchial opening was confirmed on FOB. Before repair of BEF, ventilation through tracheal lumen of DLT was avoided to avoid risk of gas insufflation into esophagus through BEF. Only right lung was ventilated with tidal volume - 300 ml, respiratory rate - 16/min, positive-end expiratory pressure - 4 mmHg, and FiO₂-1. Peak and mean airway pressure, end tidal CO₂, systemic oxygen saturation, and intermittent arterial blood gas were done during one-lung ventilation to assess adequacy of ventilation and oxygenation. Post fistula repair any difficulty in ventilation or air leak was ruled out, and thoracotomy was closed. No new tracheobronchial stent was inserted immediately after BEF repair, as there was no apparent issue with ventilation. A feeding jejunostomy was performed for enteral nutrition. The patient was awakened and his trachea was extubated soon after the surgical procedure. A thoracic epidural catheter was subsequently inserted at T4–5 intervertebral space using midline approach and bupivacaine 0.125% with 2 mcg/ml fentanyl infusion at 5–7 ml/h was used for postoperative analgesia. Parenteral paracetamol 1 g 8 hourly was used as supplemental analgesic. The postoperative recovery was uneventful, and the patient was discharged from hospital after 1 week of observation.

**Discussion**

The surgical reconstruction is considered to be the treatment of choice for tracheal stenosis resulting from prolonged endotracheal intubation or tracheostomy. Tracheal or tracheobronchial stenting is preferred technique in patients with contraindications for surgery. Stents are made up of either metal or silicone or a combination of both. Silicone stents can be easily removed postdeployment; hence, preferred in patients with reversible pathology such as stenosis. Silicone Y-stents are reserved for patients with a history of repeated migration of tracheal stent or those with bronchial pathology. In our patient silicon, tracheobronchial Y-stent was inserted due to history of migration of straight stent. Numerous complications associated with the use of stents have been reported in the literature. Martinez-Ballarin et al. found migration of stents in 17.5%, granulation tissue formation in 6.3%, and airway obstruction due to mucostasis in 6.3% of cases. Park et al. found restenosis in 40%, granulation tissue formation in 38%, migration of stents in 34%, and mucostasis in 31% of cases. The formation of tracheoesophageal fistula (TEF) has been reported after placement of esophageal stent for stricture esophagus and metallic tracheal stent for tracheal stenosis. The probable cause of fistula formation may be due to the injury at the time of stent deployment or erosion of the tracheal bronchial wall by proximal/distal margin of a malpositioned stent for prolonged time. Anesthetic management for TEF or BEF repair poses great challenge for the anesthesiologist. Early isolation of the fistula is the key in the management to prevent soiling of the healthy lung and gastric distention during positive pressure ventilation. In case with proximal TEF, this can be achieved by inserting ETT distal to TEF. The choice of right or left-sided DLT in cases of BEF depends on the side of thoracotomy approach to access the fistula. In case of thoracotomy from the diseased side, the bronchial lumen of DLT may be inserted into the opposite healthy main bronchus to ensure isolation and ventilation of healthy lung while avoiding instrumentation of fistulous bronchus [Table 1]. Alternatively, a single lumen ETT may be used on the same principle by advancing it into the desired main bronchus depending on the side of thoracotomy approach. A large more distal fistula may be occluded using Fogarty catheter. Complex airway fistulas may require bronchial intubation from surgical field or other method of ventilation such as high-frequency jet ventilation or even CPB. Our planning for the index case was based on these considerations.

Airway management of patients with tracheal stent in situ by blind intubation attempts may lead to loss of airway control due to stent displacement, dislodgement of the granulation tissue over the stent, and tracheal dissection or rupture. Bronchoscopy-guided ETT positioning may prevent these complications. In the present case, we

![Table 1: List of methods used for achieving lung isolation during repair of bronchoesophageal fistula](image)

**Table 1**: List of methods used for achieving lung isolation during repair of bronchoesophageal fistula

| Report       | Location of lesion | Surgical Approach       | Means of ventilation                          | Comment                      |
|--------------|--------------------|-------------------------|-----------------------------------------------|------------------------------|
| Giquel et al.| 1 cm above the carina | Right thoracotomy        | FOB guided left endobronchial intubation using left DLT | Left lung ventilation       |
| Patil et al. | Lower right lobe   | Right thoracotomy        | Left sided endobronchial intubation using left DLT | Left lung ventilation       |
| Ranjan et al.| Right main bronchus | Right thoracotomy        | Left sided endobronchial intubation using left DLT | Left lung ventilation       |
| Robin et al. | Distal trachea and proximal left main bronchus | Left thoracotomy | Right sided endobronchial intubation with single lumen ETT with spontaneous ventilation | Right lung ventilation     |
| Sharma et al.| Left lower lobe bronchus  | Left thoracotomy        | Right endobronchial intubation using right-sided DLT | Right lung ventilation     |

DLT: Double lumen tube, FOB: Fiberoptic bronchoscopy, ETT: Endotracheal tube
assessed the airway using flexible FOB after intubation with a small size ETT and found additional findings of tracheal mucosal bulge on the posterior side of trachea and misaligned proximal end of the stent showing only anterior margin of stent. CT scan did not infer this narrowing in the lumen of trachea and an attempt to negotiate FOB through this area into the stent failed possibly because of steep change in the direction of lumen due to posterior mucosal bulge and anterior narrowing from the misaligned proximal anterior border of the stent. This was corrected once stent was removed. This confirmed that the posterior mucosal bulge in the lumen of trachea was due to distortion of airway anatomy by malpositioned tracheobronchial stent.

To the best of our knowledge, this is the first report to show tracheal anatomical distortion and BEF formation due to malpositioned silicone tracheobronchial stent. In this case, the proximal malpositioned end caused posterior mucosal bulging and luminal narrowing of trachea whereas distal left bronchial end of the silicone stent caused formation of BEF due to pressure erosion of the left brachial and esophageal wall. The size of stent was determined based on the CT scan measurement of the tracheobronchial tree, and probably, it was appropriate or slightly larger, however; it was not properly aligned with tracheobronchial tree as apparent from the CT scan and FOB.

Conclusion

In situ malpositioned silicone, tracheobronchial stent can cause distortion of tracheal anatomy and fistula formation. Endoscopic evaluation of the tracheal lumen must be a part of airway management plan before intubation in these patients. This may reveal more information that may not be obvious from other investigation and may play a significant role in altering the management plan.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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