Case Report

Novel technique in managing bronchobiliary fistula in adults: Endobronchial embolization using silicone spigots in 2 cases

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Abstract:

Bronchobiliary fistula (BBF) can complicate most hepatic pathologies. This is a challenging group of patients, especially when surgery is precluded. The bronchoscopic application of silicon spigots is a recognized technique for the treatment of massive hemoptysis and the management of patients with bronchopleural fistula following lung resection. Their role in the treatment of BBF has never been described. In this paper we report the successful embolization using silicon spigots in two patients with BBF secondary to malignant disease, when all surgical options were exhausted.

Key words:

Bronchobiliary fistula, bilioptysis, endobronchial embolization, silicon spigot

A bronchobiliary fistula (BBF) is a communication between the biliary tract and the airway and presents with bile in expectorated bronchial secretions. It may cause cough, respiratory distress and aspiration. The etiology varies as it may be a congenital entity or a result of thoraco-abdominal trauma, biliary obstruction or hepatic infections commonly hydatid cysts or abscess. It can also be a postoperative complication of complex hepatobiliary surgery or a result of chemotherapy for liver malignancies.[1] Historically it was reported in 1850 by Peacock for patients with hydatid cysts. Their management is challenging and associated with considerable mortality. We report herein two cases treated with endobronchial embolization with silicone spigots (Watanabe spigot, EWS, Novatech, France).

Case Report

Patient 1

A 62-year-old woman presented with profuse bilioptysis after previous hepatectomy and chemotherapy for colorectal metastatic disease. Bile-leak developed in the hepato-jejunostomy which was stented through endoscopic retrograde cholangiopancreatogram (ERCP) to abolish high sphincteric pressure. The stent eroded into the duodenal wall prohibiting further stents. A hepatobiliary-iminodiacetic acid (HIDA) scan confirmed the presence of a BBF. The patient was referred to our department for palliative management. At rigid bronchoscopy, bile was leaking through the medial, lateral, and posterior segments of the right lower lobe. All segments were individually obliterated with spigots. The symptoms subsided for a month but recurred. A repeat bronchoscopy revealed leak from the remaining two basal segments which were obliterated in a similar way [Figure 1]. Successful palliation lasted for another 2 months. Thereafter in the third bronchoscopy bile was identified in the middle lobe bronchus. Two further spigots were implanted in each segment. None of the previous spigots were dislocated. The patient remained asymptomatic but succumbed from her primary disease 4 months later.

Figure 1: Bilous drainage in a segmental bronchus

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Patient 2

A 49-year-old woman presented with bilioptysis a year after hepatectomy for cholangiocarcinoma. She developed respiratory failure and required mechanical ventilation. A large hydro-pneumothorax developed in the right hemithorax with non-resolving air-leak through the chest drain. A flexible bronchoscope was passed through the endotracheal tube and bile was identified at the basal posterior segmental bronchus. A spigot was inserted and both bilioptysis and air-leak ceased allowing gradual weaning from the ventilator and resolution of the pneumothorax. Eventually she was discharged in good condition at home without any biliptysis. Five months later bilioptysis recurred. Rigid bronchoscopy revealed bile draining through the superior and lateral segmental bronchi as well as upper and middle lobe bronchi. Two spigots were used for the basal bronchi. The upper and middle lobe bronchi were not dealt with, due to high risk of patient's respiratory function compromise.

The bilioptysis was reduced significantly for a month. Then a third rigid bronchoscopy revealed bile leaking from the right anterior and medial basal segmental bronchi and additional spigots were inserted. She remained asymptomatic for a further 7 months until cancer recurrence.

Discussion

BBF seldom presents in healthy individuals. As a rule there is significant underlying hepatobiliary pathology, usually malignancy. For terminal cancer patients palliation is needed to control the significant breathing distress caused by the presence of irritating bile in the airways. As the survival averages only months it is important to ensure that patients are comfortable and symptom free for as long as possible. Benign conditions (hydatid disease) however, should be treated with curative intention.

For the nonsurgical patients the aim is decompression of the biliary tract, through ERCP or percutaneous drainage. However failed ERCPs or altered hepatobiliary anatomy after surgery may preclude such interventions.

Silicon spigots, metal plugs and endobronchial valves have all been used for bronchopleural fistulas or bronchial haemorrhage.[2-4] However bronchoscopic obliteration of a BBF has not been explored as yet. Spigots are silicon cylinders with rounded extensions to anchor in the bronchi. They are easily applicable and available in sizes of 4, 5, and 6 mm which easily fit in most segmental bronchi. A standard rigid bronchoscope with biopsy forceps is required for insertion. The second patient was intubated so flexible bronchoscope was used. In both cases the fistula tracked through more than one anatomical segment and drained in multiple segmental bronchi. Despite occluding several segments, sparing enough segments for adequate ventilation is advisable due to the poor respiratory reserves

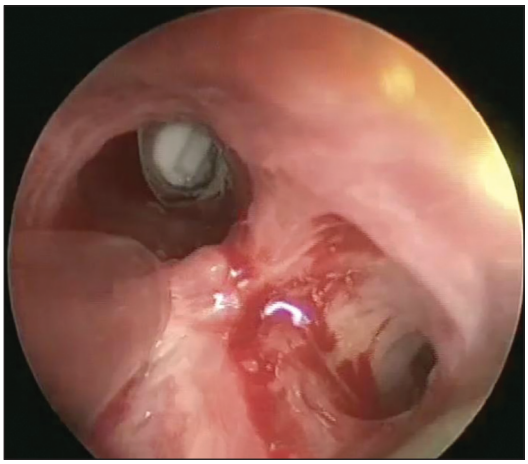

Figure 2: Occlusion of the bronchobiliary track with a silicon spigot

of such patients. Obliteration was achieved in both cases and interestingly in the second patient the concurrent air-leak ceased allowing improvement of the ventilator mechanics and extubation. A potential problem can be migration, however their small size makes them easy to expectorate. We didn't encounter any erosion. On repeat bronchoscopies the spigots remained well-seated in their positions [Figure 2]. Although both patients died of the progression of their disease, their bilioptysis was eliminated. We believe that this simple device can make a significant impact on the quality of life of patients with BBF until other suitable endoscopic applications are invented.

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