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Case Report

Endobronchial closure of broncho-biliary fistula using Amplatzer device: Case report

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ABSTRACT

Broncho-biliary fistula (BBF) is an extremely rare but serious medical condition resulting from pathological communication between the biliary system and the bronchial tree. Treatment options include both surgical and non-surgical approaches. Several endobronchial techniques, such as the spigot and glue, can be used for this purpose. This report discusses a patient who developed a broncho-biliary fistula following a liver biopsy. The BBF was diagnosed during bronchoscopy and successfully treated with an endobronchial Amplatzer device. To the best of our knowledge, this is the first report of the use of the Amplatzer device to manage BBF.

1. Introduction

Broncho-biliary fistula (BBF) is a relatively rare condition resulting from pathological communication between the biliary system and the bronchial tree and can cause severe complications.

Acquired BBF is typically caused by local infections, primary or metastatic liver tumors, biliary tract obstructions, trauma, and iatrogenic reasons [1]. In addition, invasive percutaneous procedures such as liver biopsy or ablation of hepatic lesions can contribute to developing such fistula [2].

Clinical manifestations include irritant cough, biliptysis, fever, jaundice, and abdominal or chest pain. Furthermore, a chronic course may lead to life-threatening conditions such as recurrent infections, abscesses, respiratory failure, and even death if left without proper management [1,2].

Based on the severity of symptoms, size, and location of the fistula, standard management options include either surgical or non-surgical approaches. Surgical treatment includes fistula excision, hepatectomy, or lung resection. Non-surgical management is favorable and can be divided into minimally invasive procedures (e.g., endoscopic retrograde cholangiopancreatography with biliary stenting or glue injection, percutaneous abscess drainage, bronchial occlusion) and non-invasive treatment (i.e., somatostatin and antibiotics) [3–5].

Given the development of imaging and technology, endoscopic approaches have recently gained popularity as a treatment option for various pathologies. Flexible bronchoscopy is a minimally invasive endoscopic procedure performed under moderate conscious sedation, which is particularly useful for high risk patients for anesthesia and surgery. Various bronchoscopic occluders, including covered stents, coils, spigots, biological glues, and sealants, were reported. Amplatzer devices are percutaneous, transcatheter devices of dif-

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ferent shapes, initially designed to close cardiac or vascular defects. However, albeit off-label, their use has recently gained popularity and expanded for other purposes with promising safety and efficacy results, such as bronchoscopic closure of broncho-pleural fistulas [6,7].

Herein, we describe the first successful endobronchial closure of broncho-biliary fistula using an Amplatzer device.

2. Case presentation

A 63-year-old male was diagnosed with metastatic colon cancer 11 years ago. He had undergone hemicolectomy and liver metastasectomy and subsequently received chemo- and immunotherapy. While the liver lesions had remained stable in recent years, the latest follow-up PET CT scan revealed a significant increase in their size. A lesion sample for molecular analysis was obtained through an ultrasound-guided biopsy.

Three weeks following the procedures, the patient was admitted to our hospital with cough, hemoptysis, pain in the right shoulder, and infiltrate on the right lower lobe (RLL) on the chest x-ray that did not improve after a course of oral levofloxacin. During his stay in the medical ward, he was treated with intravenous piperacillin-tazobactam for a working diagnosis of nonresolving pneumonia (shown in Fig. 1A) with parapneumonic effusion with negative bacterial cultures.

Following his discharge, the patient experienced a persistent cough accompanied by massive expectoration of bile-tinged fluid with no improvement in the RLL consolidation. He was admitted to our institute and a flexible bronchoscopy was performed. During the procedure, a significant amount of yellow-greenish fluid emanated from RLL (shown in Fig. 1B), and the broncho-biliary fistula was diagnosed. A 10 mm Amplatzer Vascular Plug was inserted into the truncus basalis bronchus with control of biliary bronchial leakage (shown in Fig. 1C, Suppl. Video 1). The patient was referred to the surgical department for drainage of the biliary tree as well as the right pleural collection (shown in Fig. 1D). After the endoscopic treatment, he reported a significant improvement in cough intensity and sputum amount.

Two weeks later, the patient was sent home with a permanent biliary drain for ambulatory follow-up (Fig. 1E).

3. Discussion

Acquired broncho-biliary fistula is a threatening condition and should be treated promptly in order to prevent serious consequences such as empyema, abscess, electrolytes loss, respiratory compromise and decrease quality of life. Although the definitive treatment is a surgical intervention, the minimally invasive percutaneous or endoscopic procedures can effectively manage BBF symptoms, promote healing, and serve as a temporary treatment before surgery. Although most of techniques that are described in literature are done by advanced gastroenterologists or interventional radiologists, several cases of endobronchial treatment of BBF were described in the past two decades. They include embolization with biological sealants and glues such as fibrin, cyanoacrylate and hyaluronic acid [8,9] or placement of silicone spigots [10–13].

While endobronchial sealants and glues can offer benefits in certain situations, they also have some potential disadvantages. An important consideration when using endobronchial glues is that their application is irreversible and cannot be adjusted after the fact. Therefore, even if the glue was initially placed correctly, migration can occur, leading to the possibility of airway obstruction. Additionally, there is insufficient information regarding these products' extended-term safety and effectiveness, including their potential degradation.

The Amplatzer devices were originally designed as a septal occluder for cardiac defects, such as patent foramen ovale. Later the device was modified as Amplatzer Vascular Plug and adapted for vascular interventions like the closure of arteriovenous fistula. It is composed of a nickel-titanium alloy (nitinol) wire mesh and can be easily inserted during an endoscopic procedure. Since its development, it has been used in several interventional non-cardiology procedures, such as interventional pulmonology, interventional radiology, and advanced gastroenterology. Its efficacy in closing broncho-pleural, gastrointestinal, and tracheoesophageal fistulas has been documented, attesting to its success in these fields [6,14,15]. Unlike silicone spigots, the shape-memory nitinol provides Amplatzer with flexibility, allowing it to conform to airway contours and be expanded or removed as needed. In addition, the device is supplied in different sizes and can be used for closure of both small and large airways.

In this article, we demonstrate a new application of the Amplatzer device as a treatment for broncho-biliary fistula with immediate symptomatic and clinical improvement without any complication.

4. Conclusion

Broncho-biliary fistula is a rare complication of liver intervention but has serious morbidity potential. Although the most common treatment is surgical intervention, there is emerging evidence of successful endoscopy management. To our best knowledge, this is the first case of broncho-biliary fistula treated by Amplatzer endobronchial insertion.

Statement of ethics

The patient provided written informed consent for publication of this case report and all the accompanying images.

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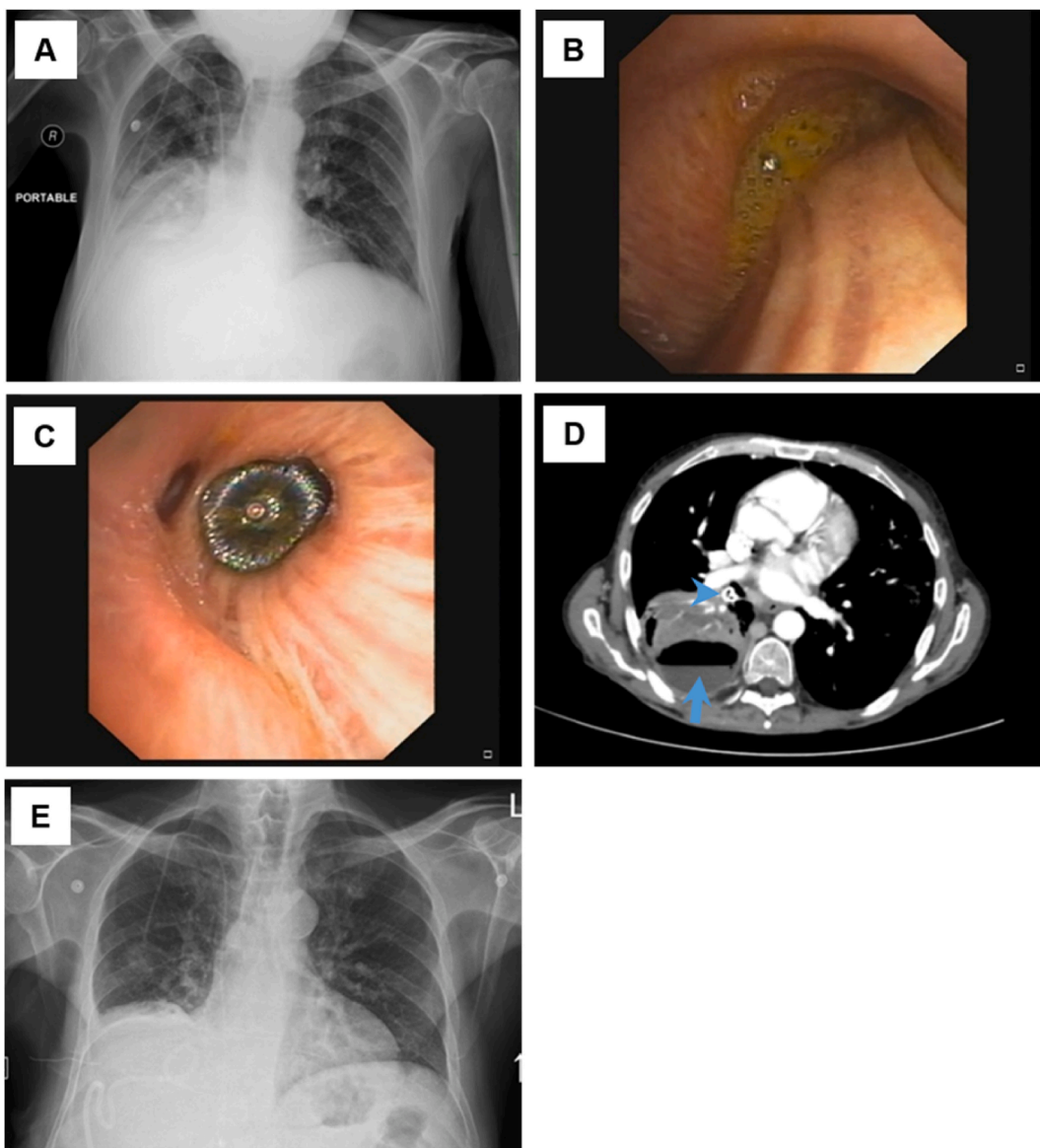


Fig. 1. Endobronchial closure of broncho-biliary fistula

A, anterior-posterior chest X-ray of the patient with right lower lobe consolidation and pleural effusion; B, endobronchial view at the level of distal trachea of biliary secretion emanating from main right bronchus during flexible bronchoscopy; C, Amplatzer device is inserted to truncus basalis and successfully occludes basal segments of the right lower lobe. Note the absence of biliary leak; D, chest CT with intravenous contrast after endobronchial treatment. Arrowhead – the Amplatzer device in right lower lobe (RLL), arrow – pleural collection with air-fluid level; E, anterior-posterior chest X-ray on follow-up visit. Note the regression of the RLL consolidation and the pig-tail catheter inserted posteriorly for drainage of the pleural collection.

Author contributions

All authors had full access to the data in the study and approved the manuscript. Procedure performance - M.R.K., Literature review - B.P., S.I., and D.R., Visualization and Writing Original Draft - L.F. and K.Z., Supervision, M.R.K

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.rmcr.2023.101943>.

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