Novel treatment (new drug/intervention; established drug/procedure in new situation)

Transhepatic embolisation of a traumatic broncho-biliary fistula: a novel approach

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Summary

Bronchobiliary fistula is a rare and challenging condition that most commonly presents worldwide following infection with hydatid cystic disease of the liver but is increasingly seen in cases of trauma involving the right upper quadrant. The most common presenting complaint is biliptysis. Treatment is initially aimed at decompressing the biliary tree which allows a considered approach for closure of the fistulous tract. Options range from conservative management to endoscopic and percutaneous approaches. Traditionally definitive treatment would have been surgical and may ultimately have resulted in hepatic and/or pulmonary segmentectomy. Current management strategies of this potentially serious condition are variable. We describe a particularly challenging case in which interventional embolisation with microcoils was used in an attempt to treat persistent post-traumatic bronchobiliary fistula in a tertiary centre. We describe this technique and hope that it may be of useful reference for those contemplating a similar approach.

BACKGROUND

A bronchobiliary fistula (BBF) is an abnormal communication between the bronchial system and the biliary tree. It can occur as a congenital malformation, although most reported cases are acquired, usually as a result of echinococcal or amoebic infection of the liver. Other causes of acquired BBF include hepatic or subphrenic abscess formation, hepatic tuberculosis, biliary lithiasis, malignancy, hepatic endometriosis, surgery, radiofrequency ablation and trauma.^{1 2} Fistulous communication between the biliary tree and thoracic structures was first described to the Royal College of Physicians by Dr Baker in 1771 remarking on 'a case of hydatids discharged by coughing, related in a letter from John Collett, M.D. Physician at Newbury, Berkshire' in a 37-year-old wife of a reverend.³

An irritant cough with biliptysis is pathognomonic of a BBF. Recurrent expectoration of bilious phlegm is characteristic of an obstruction to the normal drainage of bile, and is usually seen in noninfective causes of BBF. Chronic exposure of the bronchial tree to irritant bile causes bronchiectasis of the affected pulmonary segments. Treatment is primarily aimed at relieving the pressure of biliary obstruction allowing bile to drain into the duodenum, traditionally surgically but more recently via endoscopic retrograde cholangiopancreatography (ERCP) and biliary stenting or sphincterotomy, endoscopic ultrasound (EUS) or nasobiliary drainage.⁴

Percutaneous procedures to facilitate biliary decompression include percutaneous transhepatic biliary drainage and percutaneous balloon dilatation.^{4 5}

Depending on the underlying cause, success may be short-lived necessitating further definitive treatment aiming to close or remove the fistulous tract. Closure of the fistulous tract via bronchoscopic embolisation has been reported using acrylates.⁵ In complicated cases, or if conservative management fails, surgical intervention or

video-assisted thoracoscopic surgery may be required, and include consideration of hepatic and/or pulmonary segmentectomy.^{6 7} Aside from urgent biliary decompression to prevent the development of necrotising bronchiolitis and fulminating biliary empyema, there is no consensus as to the recommended definitive management of BBF and cases must be considered individually.^{8 9}

CASE PRESENTATION

A patient was transferred from a district hospital following a road traffic accident 2 weeks previously in which the patient sustained a grade III/IV liver injury, right pneumothorax and an inferior vena cava (IVC) injury. The patient underwent a right hemihepatectomy and IVC repair.

The patient was admitted to our tertiary hospital with symptoms of fever and malaise and subsequently had evidence of further bleeding and a significant haematoma on the right side of the abdomen and underwent an angiogram with embolisation of a pseudoaneurysm arising from a branch of the right hepatic artery adjacent to the cut surface of the liver. A few days later the patient suffered a respiratory arrest and upon transfer to the intensive care unit was noted to be coughing up large volumes of bile, and required ventilation for several days. Bile was noted to drain from the endotracheal tube.

An ERCP confirmed a BBF and a stent was placed in the lower bile duct in an attempt to encourage the bile to transit down its normal path, which had only a modest effect. The patient then represented with fever, malaise and bile expectoration and underwent a percutaneous transhepatic cholangiogram (PTC) with insertion of an internal-external drainage and fluoroscopic-guided embolisation of the BBF with coils (see technical notes, figures 1–4). The patient was discharged home 3 days later with the external drain in situ and noted to have a reduction in the biliptysis. A T-tube cholangiogram

Figure 1 Image taken from initial percutaneous transhepatic cholangiogram. Contrast injected through the needle demonstrates the biliary system. Embolisation coils from previous treatment of a right hepatic artery pseudoaneurysm. Stent in situ to allow drainage of the biliary system.

performed several days later showed normal calibre ducts with drainage demonstrated into the duodenum but no communication with the bronchial tree (figure 5). A further ERCP 1 year later again demonstrated no flow through the BBF (figure 6).

However, the patient continued to have smaller amounts of biliptysis and a bronchoscopy was performed

Figure 2 Injection of contrast through the 4F catheter clearly demonstrates flow of contrast from the right biliary system up towards the right lung, through the bronchobiliary fistula.

Figure 3 Close-up view again demonstrates the bronchobiliary fistula.

which showed bile coming from the right lower lobe bronchus. A right thoracotomy was performed via the sixth intercostal space which found the lower lobe edge of lung densely adherent to the diaphragm. This was mobilised, stapled and both sides were over sewn. Unfortunately, this did not resolve her biliptysis and a CT cholangiogram was performed which showed high-density material arising from the posterior right surface of

Figure 4 Several vortex coils are placed along the bronchobiliary fistula, above the previous coils, obstructing the track. An internal/external drain is placed at the same time in order to divert the flow of bile.

Figure 5 Follow-up tubogram 1 week later demonstrates no flow of contrast through the bronchobiliary fistula.

the convexity of the liver extending into a right-sided posteriorly situated pleural collection. It was felt that the area of remaining right lobe of liver was not draining into the normal biliary system and was continuing to drive the fistula. A completion right hemihepatectomy was performed which appeared to resolve the symptoms. However, the patient experienced recurrence of biliptysis 6 months later and an MRI was performed that suggested a small area of un-drained liver in segment 4 that was likely causing the ongoing biliptysis. Further surgery was

Figure 6 Image taken from endoscopic retrograde cholangiopancreatography 1 year after embolisation of the bronchobiliary fistula (BBF). Again there is no flow of contrast through the BBF demonstrated.

performed and the fistula across the diaphragm was found, taken down and closed. Diseased remnants of the liver were removed and a leak located in the right bile duct was secured. One year on the patient has not had any further episodes of biliptysis and has regained weight.

INVESTIGATIONS

The imaging modalities used in diagnosis of BBF include US, PTC, CT, hepatobiliary scintigraphy, single-photon emission CT (hybrid SPECT/CT), MRI and MR cholangiopancreatography.¹⁰

TREATMENT

The interventional radiology-guided embolisation procedure was performed under sedation and local anaesthetic, 10 ml of Lidocaine and intravenous midazolam and morphine were given until the patient was lightly sedated and comfortable.

Ultrasound confirmed a nondilated biliary tree and a puncture of the left hepatic duct was performed using a 21G Accustick needle (Boston Scientific). Access into the left biliary duct was obtained with a 0.18 wire and a 4F catheter. Position was confirmed with injection of contrast (Visipaque 320) under fluoroscopy. A 7F vascular sheath Cordis was positioned to secure access to the biliary system. Initial cholangiogram confirmed a leak of contrast from segment 4 duct with clear passage of contrast via the liver parenchyma into the bronchial tree confirming the fistula (figures 1–3)

A 4F (COOK) Biliary Manipulation Catheter (BMC) and a guide wire were used to negotiate into the common bile duct. The previous ERCP plastic stent was noted to be lying across the ampulla. Alongside the wire a Cordis Multi Purpose Catheter was used to negotiate a 0.35 angled Terumo wire into the duct that was clearly leaking. The track was again demonstrated and embolisation of this track was then performed using multiple vortex coils (COOK), the actual segmental duct was also coiled at this stage (figure 4). Cholangiogram at the end of the procedure confirmed that there was no leakage of contrast via the track into the bronchus.

A 32 sidehole 8.5F COOK locking Pigtail drain was left on internal external drainage from the left duct access site into the duodenum to allow the bile to bypass the embolised tract. This was secured with a 2/0 silk suture. There were no immediate complications postprocedure.

A follow-up cholangiogram was performed 1 week after this procedure to ensure that tract remained occluded and that the drain could be removed safely. This confirmed that the biliary ducts were of normal calibre with drainage into the duodenum demonstrated. There was no communication with the bronchial tree noted (figure 5).

DISCUSSION

This report describes the novel approach of using coil embolisation in the treatment of a BBF track in an attempt to both reduce the flow along the fistula and serve as a form of mechanical obstruction. To our knowledge this is the first case describing this approach. We found that this technique can be combined at the same time as insertion of an internal/external biliary drain to

further divert the flow of bile away from the BBF to maximise the effect.

However, this method may not be the definitive treatment for BBF. It nonetheless is a valuable adjunct to endoscopic or bronchoscopic intervention in the management of BBFs, in appropriate patients. Selective embolisation of the fistulous track is safe and may decrease the morbidity associated with the condition and may avert or delay the need for surgery until a more suitable opportunity in patients presenting with a BBF. In the complex case we describe there was obstruction of the BBF more than 1 year after the deployment of the coils, as confirmed by the reduction in the patient's symptoms and the follow-up tubogram and ERCP that were performed. Although the main fistula track was obliterated by the coils, smaller tracks may not have been obstructed by the coils, due to their size, and this resulted in ongoing symptoms which eventually led to the patient requiring surgery to excise the residual BBF.

Although not performed in this case, the use of gel foam along with microcoils may be an alternative approach that may accentuate the mechanical obstruction by obliterating any smaller channels. An important characteristic of any embolic agent used in the biliary system is its ability to be used in a controllable manner and therefore coils may be the first choice material. Other embolic material that may be helpful such as onyx or histacryl may be used but are potentially less controllable and therefore their use may be limited. Further evidence and studies of the long-term outcome of the use of these agents in the biliary tree is required.

Learning points/take home messages

- ▶ A traumatic broncho-biliary fistula is a rare finding and should be suspected in a patient presenting with biliptysis following trauma.
- ▶ Embolisation of the broncho-biliary fistula is technically possible and may reduce the morbidity in these patients and delay/avert surgery.
- ▶ This remains a novel approach in treating these patients.

Competing interests None.

Patient consent Obtained.

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