On-line Case Report

Cholecystobronchial fistula secondary to adenomyomatosis of the gallbladder

CD MANN¹, NA JOHNSON¹, MS METCALFE¹, CP NEAL¹, RF HARRISON², DP BERRY¹, AR DENNISON¹

Departments of ¹Hepatobiliary and Pancreatic Surgery and ²Pathology, University Hospitals of Leicester, Leicester General Hospital, Leicester, UK

ABSTRACT

A case of cholecystobronchial fistula secondary to adenomyomatosis of the gallbladder is described. A cholecystobronchial fistula is a very unusual cause of fistulation between the bronchial and biliary tree. This is only the fifth reported case in the English language literature.

Keywords: Cholecystobronchial fistula – Adenomyomatosis – Gallbladder

A 79-year-old man, normally fit and well, presented with a 5-week history of mild dyspnoea and a cough productive of copious amounts of dark-yellow sputum, which had not responded to oral antibiotic treatment prescribed by his general practitioner. He complained of no other symptoms and had an unremarkable past medical history apart from well-controlled epilepsy. He was a non-smoker and consumed only a small amount of alcohol.

On examination, the patient was pyrexial at 38.5°C, tachycardic, and tachypnoeic, with oxygen saturations of 95% on room air. Examination of his respiratory system demonstrated reduced air entry and dullness to percussion at the right lung base. Abdominal examination was unremarkable and he was not jaundiced. Blood tests on admission revealed a neutrophilia (white cell count $21 \times 10^9/l$; neutrophils $17.53 \times 10^9/l$), but no other abnormalities. A chest X-ray demonstrated an elevated right hemidiaphragm, a right-sided pleural effusion and an area of consolidation above. The initial working diagnosis was pneumonia and the patient was treated with appropriate

antibiotics. Microbiological examination of the sputum demonstrated coliform bacilli, and this, in addition to the colour of the sputum, raised concerns about its origin. Sputum analysis demonstrated the presence of bilirubin and suggested a bronchobiliary fistula.

An MRCP demonstrated a 10 x 5 cm multiloculated, right subphrenic collection and a small right-sided pleural effusion. There were calculi in the gallbladder, but no dilated ducts or suggestion of a fistula. A hepatobiliary iminodiacetic acid (HIDA) scan also failed to demonstrate a biliary leak. An ERCP was performed, which demonstrated no hepatic duct dilation or obstruction. Leakage of contrast was noted from the right hepatic lobe superiorly towards the subphrenic collection found on MRCP, and a biliary stent was inserted to improve biliary drainage. In an attempt to explain the origin of the fistula, serum tumour marker tests were performed, but these were unremarkable. The biliopsy, however, did not settle after ERCP; therefore, a laparoscopy was performed and the subphrenic collection

Correspondence to: Christopher D Mann, Clinical Research Fellow, Department of Hepatobiliary and Pancreatic Surgery, Leicester General Hospital, Gwendolen Road, Leicester LE5 4PW, UK

T: +44 (0)116 2490490 Ext 8110; M: +44 (0)7734 203808; F: +44 (0)116 2584708; E: chris.mann@doctors.org.uk

Figure 1 Histological examination of gallbladder fundus. A nodule of adenomyomatosis demonstrating invagination of mucosa into the smooth muscle of the muscularis layer, forming several large Rokitsky-Aschoff sinuses (low power magnification).

drained. An intra-operative ultrasound, limited due to adhesions, did not identify any other sinister pathology or a defect in the diaphragm. Clinical response was dramatic, the biliptysis settling immediately, and the patient was discharged with drains *in situ*. Review at one month was notable for complete cessation of biliptysis and minimal production from the abdominal drains, which were removed.

Unfortunately, the patient re-presented 2 months later with recurrent biliptysis. A repeat CT was performed which demonstrated bowel adherent to the superior surface of the liver and a defect in the diaphragm with adjacent consolidation. Air was also noted in the biliary tree. Due to failed conservative treatment, it was decided that formal operative closure of the fistula was appropriate. At laparotomy, after the extensive adhesions had been cleared, the gallbladder was found to be elevated in a position similar to that achieved at laparoscopic cholecystectomy with the fundus of the gallbladder adherent to the diaphragm. A fistula was found connecting the fundus to a hole in the right central diaphragm. The fistula was disconnected, a cholecystectomy performed, and the diaphragmatic defect repaired. A t-tube was inserted into the bile duct, and both abdominal and transabdominal chest drains inserted. In addition, thorough bronchial lavage was performed by bronchoscopy. Culture of the bile grew coliform bacilli and *Enterococcus faecalis*, which were treated with vancomycin and imipenem. The histology of the gallbladder demonstrated focal adenomyomatosis at the fundus of the gallbladder, with a fistula tract through a Rokitsky-Aschoff sinus (Fig. 1).

The patient spent 24 h in the high dependency unit and made a good postoperative recovery on the ward. He was discharged following complete resolution of the biliptysis 2 weeks after laparotomy. Follow-up hepatic ultrasound and t-tube cholangiogram were unremarkable and the t-tube removed 3 weeks following discharge. The patient is currently well and free of biliptysis 5 months following laparotomy.

Discussion

Bronchobiliary fistula is an uncommon condition. Fistulation usually results from an obstructed biliary tree or trauma (both surgical and non-surgical). Other published causes include hydatid disease, hepatic abscesses, intrahepatic malignancy, and congenital bronchobiliary fistula. A cholecystobronchial fistula is a very unusual cause of fistulation between the bronchial and biliary tree. This is only the fifth reported case in the English language literature;¹⁻⁴ as far as the authors are aware, this is the first reported case of cholecystobronchial fistula secondary to adenomyomatosis of the gallbladder. Adenomyomatosis (also known as cholecystitis glandularis proliferans and diverticulosis of the gallbladder) is identified in approximately 5% of specimens obtained at cholecystectomy. It is an acquired disease of unknown aetiology, although it has been suggested that it may be a degenerative condition secondary to high intraluminal pressures. Adenomyomatosis is generally considered a benign condition, although there have been reports of carcinoma occurring in localised areas of adenomyomatosis.⁵ Pathologically, adenomyomatosis is characterised by hyperplasia of the gallbladder glandular mucosa and hypertrophy of the smooth muscle, combined with variable inflammatory and fibrotic changes. Invagination of the glandular mucosa through the muscularis forms Rokitsky-Aschoff sinuses, which are pathognomonic for the diagnosis of adenomyomatosis. In this way, adenomyomatosis is considered analogous to diverticular disease of the colon. The majority of patients with adenomyomatosis also have gallstones; consequently, the diagnosis is usually made incidentally on histology after cholecystectomy. Most patients with acalculous adenomyomatosis remain asymptomatic and it is likely that in cases associated with calculi, it is the gallstones themselves responsible for the symptoms. Although there is no clear relationship between adenomyomatosis and gallstones, it is possible that gallstones, particularly if associated with chronic inflammation, may result in high intraluminal pressure predisposing to the condition. In this patient, it is unclear how this resulted in fistula formation. If considered analogous to diverticular disease of the colon, a perforation of a Rokitsky-Aschoff

sinus could have formed a localised abscess and subsequent fistulation through the diaphragm, resulting in the cholecystobronchial fistula.

The initial management of a suspected bronchobiliary or cholecystobronchial fistula should ensure free biliary drainage, in this case via stenting at ERCP, although percutaneous biliary drainage remains an option. In addition, if an abscess cavity is present, this should be drained, either percutaneously by ultrasound or CT-guidance, or laparoscopically. In the course of investigation, it is always necessary to exclude any underlying malignant disease as a cause of the fistulation; in this case, no parenchymal lesion was identified on CT, MRCP or laparoscopic ultrasonography, and serum tumour markers were normal. If these measures succeed and the biliptysis resolves, in the case of cholecystobronchial fistula, formal exploration and cholecystectomy may be performed at a later date. This allows any procedure, which may be extensive, to be performed on an elective

basis. Unfortunately, in this case, conservative measures failed and early formal operative closure was required.

References

1. Senatore CM, Collarini HA, Navarret EE. Biliobronchial fistula; regarding a case of cholecystobronchial communication. *Prensa Med Argent* 1958; **45**: 485–8.
2. Delco F, Domenighetti G, Kauzlaric D, Donati D, Mombelli G. Spontaneous biliothorax (thoracobilia) following cholecystopleural fistula presenting as an acute respiratory insufficiency. Successful removal of gallstones from the pleural space. *Chest* 1994; **106**: 961–3.
3. Allison MC, Milkens S, Burroughs AK, Rogers HS, Thomas HC. Bronchobiliary fistula due to acute cholecystitis in a suprahepatic gallbladder. *Postgrad Med J* 1987; **63**: 291–4.
4. Pogany K, van Bentam N, Koch P, de Bruin W, Kolkman J. Cholecystobronchial fistula. *Endoscopy* 2000; **32**: S38.
5. Nabatame N, Shirai Y, Nishimura A, Yokoyama N, Wakai T, Hatakeyama K. High risk of gallbladder carcinoma in elderly patients with segmental adenomyomatosis of the gallbladder. *J Exp Clin Cancer Res* 2004; **23**: 593–8.