CASE REPORT

Choledochoduodenal fistula: an unusual case of pneumobilia

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SUMMARY

Pneumobilia is the finding of air in the biliary tree. Most cases are iatrogenic in origin, especially after sphincterotomy and after hepaticojejunostomy or choledochojejunostomy. In patients without such history, the presence of pneumobilia needs further investigation. Most patients are likely to have an enterobiliary fistula. Although patients may be asymptomatic, possible complications include gallstone ileus, Bouveret syndrome or recurrent episodes of cholangitis. We present a case of a 38-year-old man presenting with obstructive jaundice and pneumobilia in whom choledochoduodenal fistula was diagnosed at endoscopic retrograde cholangiography. A description of different types of choledochoduodenal fistulas and management recommendations are also provided.

BACKGROUND

Enterobiliary fistula was first described by Bartholin in 1654.¹ Sixty-eight per cent of cases occur between the gallbladder wall and duodenum. This may cause the well-known gallstone ileus and Bouveret syndromes. Only 8.6% of enterobiliary fistulas are of the choledochoduodenal type.² This type of fistula may present with symptoms of peptic ulcer disease or as recurrent episodes of cholangitis. A high index of suspicion is needed for the correct diagnosis. Here we present a case of choledochoduodenal fistula in order to raise awareness of this rare cause of pneumobilia.

CASE PRESENTATION

A 38-year-old man, a new immigrant from Ethiopia, presented at our institution's emergency department with epigastric pain for 5 years and a 2-month history of nausea, vomiting and pale stool. He was otherwise healthy. This was his first presentation.

On physical examination, the patient was afebrile and mildly jaundiced. There was mild tenderness in the epigastrium.

Laboratory tests revealed leucocytosis of 11 900/ μ L with 86% neutrophils and C reactive protein of 183 mg/L. Transaminases were slightly elevated (aspartate aminotransferase 62 IU/L, alanine transaminase 125 IU/L), total bilirubin at presentation was 4.4 mg/dL and direct bilirubin 2.2 mg/dL, alkaline phosphatase 260 IU/L γ -glutamyl transpeptidase 239 IU/L, amylase 45 IU/L and cancer antigen 19-9 was elevated at 140 U/mL (range 0–39).

INVESTIGATIONS

Abdominal X-ray revealed air within the biliary tree (figure 1). On abdominal ultrasound there was

Figure 1 Plain abdominal X-ray showing air in biliary tree (white arrow).

thickening of the gallbladder wall and pericholecystic fluid, but no sludge or stones. The intrahepatic bile ducts were dilated and the common bile duct (CBD) measured 10 mm in width, raising suspicion of a distal obstruction. Pneumobilia was confirmed.

CT showed a 4 cm gallbladder with wall thickening and without cholelithiasis. The CBD was 14 mm wide with hyperdense material at its distal end (figure 2). The intrahepatic biliary tree was enlarged bilaterally with pneumobilia (figure 3). The pancreas was normal.

Although the cause of pneumobilia remained unclear, radiological findings suggested cholecystitis and choledocholithiasis.

Figure 2 CT showing gallbladder with wall thickening (white arrow) and enlarged common bile duct with sludge (black arrow).

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Figure 3 CT showing air within intrahepatic bile ducts (white arrows).

DIFFERENTIAL DIAGNOSIS

The most common cause of pneumobilia is iatrogenic—after endoscopic retrograde cholangiopancreatogram (ERCP) with or without sphincterotomy or after hepaticojejunostomy or choledochojejunostomy. Other cases include enterobiliary or broncho-biliary fistula, emphysematous cholecystitis or liver abscess, blunt abdominal trauma or cardiopulmonary resuscitation and congenital incompetence of the sphincter of Oddi.

TREATMENT

The patient was admitted for intravenous antibiotic therapy and an ERCP was planned to elucidate a diagnosis and relieve jaundice.

At ERCP the major duodenal papilla was normal in appearance but the injected contrast fluid returned to the duodenum via a suprapapillary choledochoduodenal fistula. After papillotomy, biliary sludge was extracted with a basket sweep.

OUTCOME AND FOLLOW-UP

After ERCP the patient's symptoms were relieved and liver function tests returned to normal. The patient was discharged. Ambulatory MR cholangiopancreatogram (MRCP) showed a normal CBD with no stones. There were no stones in the gallbladder. No fistula was seen. The decision was taken, therefore, to manage the patient conservatively. The patient remains asymptomatic after 1 year follow-up.

DISCUSSION

The gallbladder is in close apposition to its surrounding organs. The inferior surface of the fundus and the body of the gallbladder lies close to the transverse colon. The neck of the gallbladder and the cystic duct lie on the first part of the duodenum. The CBD descends in the free edge of lesser omentum behind the first

Figure 4 Barium swallow test demonstrating reflux of contrast into the common bile duct. Adapted from Hoppenstein *et al.*⁵

part of the duodenum, approximately 7 cm distal to the pylorus, and opens into the major duodenal papilla.

Of all the types of enterobiliary fistula, the most common is cholecystoduodenal (table 1).

This condition is generally caused by a stone impacted at the neck of the gallbladder with secondary pressure necrosis of the gallbladder wall and fistula formation into the first part of the duodenum.

Choledochoduodenal fistula (CDF) may occur at two distinct sites in the CBD, and is described in terms of two broad classes (proximal and distal). Since the pathological cause is different in each type, it is important to distinguish between them in order to manage treatment accordingly.

Proximal CDF, the most common type, occurs between the CBD and posterior wall of the duodenal bulb. In 80% of cases

Figure 5 Gastroduodenoscopy showing ostium with bilious secretion entering through the posterior wall of the duodenal bulb. Adapted from Neumann *et al.*⁶

Table 1 Types of enterobiliary fistula²

Type of enterobiliary fistula	Percentage of cases
Cholecystoduodenal	68
Cholecystocolonic	13.6
Choledochoduodenal	8.6
Cholecystogastric	4.9
Duodeno-left hepatic	4.9

Figure 6 Endoscopic repair of a proximal choledochoduodenal fistula with an 11 mm over-the-scope clip. Adapted from Neumann *et al.*⁶

there is a history of long-standing dyspepsia and the pathophysiology is of acid-dependent duodenal ulceration and fistulation into the posteriorly located proximal CBD. Rarely, it can present as upper gastrointestinal bleeding, sometimes massive, because of ulceration and perforation into the gastroduodenal or superior pancreaticoduodenal artery.³ Kyle⁴ described the sudden relief of dyspeptic symptoms after such fistula formation because of the alkalisating property of bile relative to the hyperacid environment of the ulcer. Other causes are secondary to duodenal diverticulitis, paraduodenal abscess, trauma and carcinoma of the duodenum or bile duct. Barium swallow demonstrates reflux of contrast into the bile ducts (figure 4).⁵ The diagnosis is made at endoscopy when bile is seen entering through the posterior wall of the duodenal bulb. (figure 5).⁶ Ulcer treatment with a proton-pump inhibitor or a H₂ antagonist is sufficient in the treatment of this type of fistula.⁷ For refractory cases, vagotomy with distal gastrectomy (antrectomy) and gastrojejunostomy with Billroth II reconstruction has been suggested, leaving the fistula intact.⁸ Closure of the fistula with a covered metal stent has been recently described⁹ (figure 6).⁶

Distal CDF occurs near the entrance of the distal CBD into the second part of the duodenum. Typically, in 90% of cases, the cause is an impacted stone in the distal 2 cm of the CBD, producing erosion of the CBD wall and fistulation into the descending

Figure 7 Ikeda classification of distal choledochoduodenal fistula. CBD, common bile duct; MPD, main pancreatic duct; P, papilla of Vater; F, fistula.¹¹

duodenum.¹⁰ Other causes are ampullary carcinoma or cholangiocarcinoma of the distal CBD. Patients usually present with the classic triad of cholangitis: right-upper quadrant pain, fever and jaundice. Ikeda and Okada¹¹ further classified distal CDF into type I and type II (figure 7). In type I the fistula arises on the longitudinal fold just adjacent to the papilla as a result of a small stone impacted at the intramural portion of the CBD. In type II, named suprapapillary distal CDF, as in our case, the fistula arises on the duodenal mucosa itself, proximal but adjacent to the duodenal fold. In this type of CDF, gallstones impacted are typically larger and impact in the extramural portion of CBD. Diagnosis of distal CDF is made at ERCP with back-flow of the injected contrast from the fistula itself (figure 8A). Treatment is typically carried out during ERCP with fistulotomy, extended sphincterotomy and extraction of stones¹² (figure 8B). If not already performed, laparoscopic cholecystectomy should later be performed. For higher fistula within the distal CBD when fistulotomy is not safe, choledochojejunostomy is a surgical option.¹³

Figure 8 Photographs from endoscopic retrograde cholangiopancreatogram. (A) The ampulla of Vater with cannula in place. Contrast injected through the cannula flows back into the duodenum through the fistula tract. (B) After papillotomy, two large common bile duct stones being extracted into the duodenum using the papillotomy. Adapted from <http://www.EndoAtlas.com>, Atlanta South Gastroenterology.

Learning points

- In patients with peptic ulcer disease and pneumobilia, one must consider the presence of a proximal choledochoduodenal fistula. Standard medical treatment of peptic ulcer disease will heal the fistula.
- Patients with recurrent episodes of cholangitis should be investigated for a distal choledochoduodenal fistula, and endoscopic retrograde cholangiopancreatogram performed for diagnosis and extended sphincterotomy.
- A high index of suspicion is needed for this relatively easy-to-treat condition.

Competing interests None.

Patient consent Obtained.

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