

# Pneumobilia with Cholangitic Abscess: A Case Report

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## Abstract

Pneumobilia or aerobilia is an accumulation of air in the biliary tree. Pneumobilia is considered as a serious pathology, requiring urgent surgical intervention. Most cases are iatrogenic in origin, occurring post-surgeries for sphincterotomy or hepaticojejunostomy, or choledochojejunostomy. We report here a case of 32-year-old woman presenting with high fever and abdominal pain with air in the biliary tree and cholangitic abscess, which did not subside in spite of being on full dose higher antibiotics. Eosophagogastroduodenoscopy revealed multiple choledochooduodenal fistulas. Magnetic resonance cholangiopancreatography confirmed pneumobilia. The patient was advised 4-weeks rest, after which surgical intervention would be contemplated and followed up with ultrasonography scan weekly.

**Key words:** Aerobilia, Biliary tree, Cholangitic abscess, Choledochooduodenal fistula, Pneumobilia

## INTRODUCTION

Pneumobilia or aerobilia is an accumulation of air in the biliary tree.<sup>1</sup> The most common conditions associated with pneumobilia include: (1) A biliary-enteric surgical anastomosis, (2) an incompetent sphincter of Oddi, or (3) a spontaneous biliary-enteric fistula, and rarely. (4) Biliary bronchopleural fistula.<sup>2</sup> Most cases of pneumobilia are related to gallstone disease. Pneumobilia is considered as a serious pathology, requiring urgent surgical intervention.

We report here a case of 32-year-old woman presenting with high fever and abdominal pain with air in the biliary tree and cholangitic abscess.

## CASE REPORT

The 32-year-old woman was brought by her husband with a history of fever, bilious vomiting, and mild abdominal pain since 5 days. Fever was high grade associated with chills. The patient had also had nausea with 2-3 episodes of bilious vomiting, and mild abdominal pain. The patient

had undergone cholecystectomy 20 years back in view of congenital choledochal cyst of the common biliary duct (CBD); the post-operative period until now was uneventful. No other significant past medical/surgical history was elicited. Her abdominal examination revealed severe tenderness in the right hypochondriac region. Other systemic examination revealed no abnormality.

Abdominal ultrasound showed mild fullness of intrahepatic biliary radicals (IHBR) in the right lobe of the liver with few echogenic foci within. Computed tomography scan of the abdomen was performed which showed dilatation of the central and peripheral IHBR with pneumobilia, and multiple calculi in these IHBR. Few round cystic lesions, likely cholangitic abscesses, could be seen in the peripheral aspects of segment VII of the right lobe of liver communicating with the biliary radicals. Eosophagogastroduodenoscopy (OGDscopy) showed multiple fistulous openings at D1-D2 junction, considered to be choledochooduodenal fistulas. Magnetic resonance cholangiopancreatography (MRCP) was advised for further confirmation. The MRCP confirmed the presence of air foci in the proximal CBD, representing choledochooduodenal fistula, with dilatation of central and peripheral IHBR, with pneumobilia, with multiple calculi in the dilated IHBR (Figures 1-4).

## Management

The patients' blood was investigated and revealed her hemoglobin (Hb) to be 10.8 mg/dl, total leukocyte count - 14000/ $\mu$ l, and platelets of 314 $\times$ 10<sup>3</sup>/ $\mu$ l.

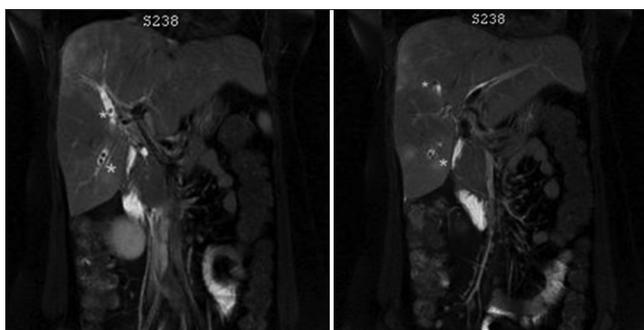
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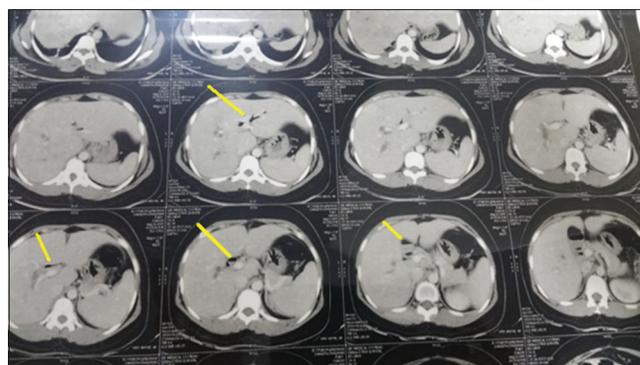
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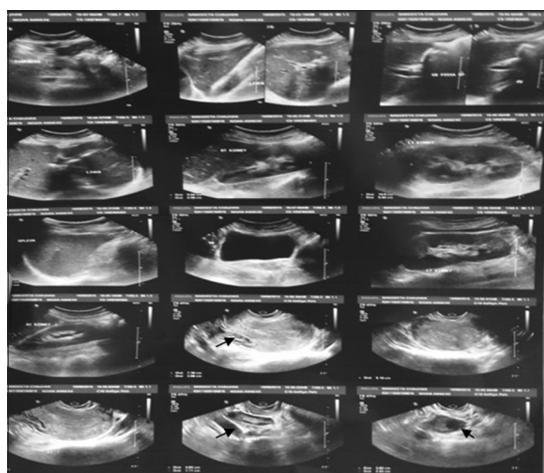
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**Figure 1: Coronal T2-weighted (T2W) magnetic resonance (MR) image reveals that the suspected stones are air bubbles (\*) within the right hepatic biliary radical. MR cholangiopancreatography images may be misinterpreted therefore T2W images are most significant in identifying the pneumobilia**



**Figure 3: Abdominal computed tomography-scan. Dilatation of central and peripheral intrahepatic biliary radicals (IHBR) with pneumobilia, and multiple calculi in these IHBR. Few round cystic lesions, likely cholangitic abscesses could be seen in the peripheral aspects of segment vii of right lobe of liver communicating with the biliary radicals**



**Figure 2: Abdominal ultrasound. Arrows represent mild fullness of intrahepatic biliary radicals in the right lobe of liver with few echogenic foci within**



**Figure 4: Oesophagogastroduodenoscopy. Fistulous openings at D1-D2 junction, considered to be choledocho-duodenal fistulas**

In spite of starting the patient on higher antibiotics in full doses - injection piperacillin, injection metronidazole, the fever and abdominal pain did not subside. OGDscopy was done; which revealed multiple choledochoduodenal fistulas and was advised MRCP for further evaluation. The presence of pneumobilia was confirmed by MRCP, after which she was advised rest for 4 weeks till sepsis settled, after which surgical intervention would be contemplated and was asked to follow-up with ultrasonography scan weekly.

## DISCUSSION

Pneumobilia is the presence of air in the biliary tree. Most cases are iatrogenic in origin, occurring post-surgeries for sphincterotomy or hepaticojejunostomy, or choledochojejunostomy. In the absence of a history of these procedures, other causes should be looked for. Mostly,

such patients do have enterobiliary fistula of some origin, or more ominous, cholangitis. Spontaneous pneumobilia may occur due to gallstone erosion in the biliary tree to an adjacent viscus.<sup>3</sup>

Our patient was a 32-year-old woman with a history of congenital choledochal cyst of CBD, followed by cholecystectomy, 20 years ago. Her post-operative period was uneventful. However, her present imaging investigations revealed multiple calculi in the IHBDs, air foci in the proximal CBD, and intrahepatic cholangitic abscesses.

Pneumobilia has been reported to have a presentation similar to that of cholelithiasis. A long-standing history of biliary stones, recurrent biliary tract infections, and the presence of CBD stones do predispose the occurrence of choledochoduodenal fistula when seen with cholelithiasis.<sup>4</sup> Previous biliary surgery is a lesser contributing factor, as seems to be the case in our report. Choledochoduodenal fistula leads to exposure of biliary tract to gut flora. This not only complicates the case by the occurrence of

cholangitic abscess, but also leads to biliary diarrhea, fluid and electrolyte wasting, and malabsorption.

Abdominal computed tomography (CT) is a valuable diagnostic method as fistulas, air in biliary tree, in addition to contraction of the gallbladder, can be visualized. Although our patient was cholecystectomized, CT imaging of a biliary enteric fistula is also useful for differentiating between a gall bladder enteric fistula and a common bile duct-enteric fistula.<sup>5</sup>

Proximal choledochoduodenal fistulae are managed surgically. Two main surgical approaches, one-stage procedure (comprising enterotomy, cholecystectomy, and fistula repair) and two-stage procedure (enterotomy with or without subsequent cholecystectomy and fistula repair) are available.<sup>6</sup> There is no consensus on which is better. In our case, the patient being previously cholecystectomized was managed with high dose antibiotics and OGDscopy done initially on presentation and revealed multiple choledochoduodenal fistulas. CT-abdomen was done which was suggestive of dilatation of central and peripheral IHBR with pneumobilia and findings of the cholangitic abscess. MRCP was advised which confirmed the presence of air foci in the proximal CBD, representing

choledochoduodenal fistula, with dilatation of central and peripheral IHBR, with pneumobilia, with multiple calculi in the dilated IHBR.

## CONCLUSION

Pneumobilia is a rare entity. Occurring with cholangitic abscess in a patient with an uneventful cholecystectomy, it presents a challenging case for determining the diagnosis as well as the management.

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