Low convergence of hepatic ducts: A rare extrahepatic biliary tree anatomical variation

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ABSTRACT

Introduction: Low convergence of hepatic ducts is extremely rare. Here, the right and the left hepatic ducts course down towards the second part of the duodenum separately and converge just proximal to the ampulla of Vater, forming a short common bile duct. Case Series: We describe 2 cases of a 43-year-old male and a 53-year-old female with this anatomical anomaly who presented with obstructive jaundice. Endoscopic retrograde cholangiopancreatography (ERCP) demonstrated low convergence of the hepatic ducts. Both cases were associated with biliary ectasia and complicated with primary ductal stones. Excision of the anomalous extrahepatic bile ducts with hepaticoenterostomy was performed for one of the cases, however the other was lost to follow-up. Conclusion: Definitive operative intervention seems prudent despite the lack of consensus in the management of such anomaly, especially when there are concomitant biliary ectasia and ductal stones.

Keywords: Biliary stone disease, Distal bifurcation, Extrahepatic biliary tract, Hepatic ducts, Low convergence

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INTRODUCTION

Anatomical variations of the biliary tree are not uncommon and numerous anomalies have been described in literature. The right and the left hepatic ducts converge to form the common hepatic duct just after exiting the liver hilum in 60–70% of the cases [1]. However, low convergence of the hepatic ducts is extremely rare and the exact incidence is unknown with review of literature resulted in only a few case reports [2, 3]. Here, the right and the left hepatic ducts course down towards the second part of the duodenum separately and converge just proximal to the ampulla of Vater, forming a short common bile duct. This may become significant
during operative intervention as failure to recognize such anomaly may lead to iatrogenic biliary injury.

CASE SERIES

**Case 1:** A 43-year-old male was presented with epigastric pain and obstructive jaundice. Ultrasonography (USG) showed a normal liver and gallbladder with dilated common bile duct (CBD) and intrahepatic ducts (IHD) secondary to choledocholithiasis. Endoscopic retrograde cholangiopancreatography (ERCP) confirmed these findings (Figure 1). The ERCP with attempted stone extraction was challenging, made difficult by the presence of a huge pre-pyloric gastric ulcer. Thus a biliary stent was inserted to temporarily relieve the obstruction. He underwent open cholecystectomy and CBD exploration. Intraoperatively, the CBD was dilated with multiple soft stones within. Cholangioscopy confirmed no residual stones. Intraoperative biliary stenting was done. He recovered uneventfully and the biliary stents were removed two months later. Endoscopic retrograde cholangiopancreatography revealed the presence of low convergence of the hepatic ducts (Figure 2). The patient was counseled for operative intervention but was not keen and subsequently was lost to follow-up.

**Case 2:** A 53-year-old female who had previous open cholecystectomy for empyema of the gallbladder 16 years ago presented with epigastric pain and obstructive jaundice. The ERCP revealed low convergence of the hepatic ducts with a large stone in the left hepatic duct (Figure 3). Both the hepatic ducts and the CBD were grossly dilated and ectatic necessitating biliary stenting. Computed tomography (CT) scan showed normal liver, mild dilatation of the IHD with marked dilatation of the left and right hepatic ducts and CBD. Cholangiojejunostomy was performed and intraoperatively, both the hepatic ducts and CBD were dilated with 1 large stone in the left hepatic duct, measuring 2.5x1.5 cm and 2 smaller stones in the right hepatic duct, measuring 1.0x1.0 cm. Both hepatic ducts were transected as proximal as possible close to the lower liver edge. The medial edges of the ducts were sutured with interrupted polyglyconate sutures MaxonTM 4/0 to fashion a single duct and a retrocolic cholangiojejunostomy was created (Figure 4 and Figure 5). Histopathological examination of the hepatic ducts and CBD showed chronic inflammation. Subsequent follow-ups found she was well with no stone recurrence.

DISCUSSION

Anatomical variations of the biliary tree are not uncommon and numerous anomalies have been described in literature, based on observation from imaging studies, operative reports and autopsy discoveries, in as high as 47% of the cases [4]. These anomalies may be minor or major with variable clinical significance.
The numerous anatomical variations and anomalies are attributable to the underlying complexity of the embryological development of the liver and biliary tree. By the fifth week of intrauterine life, the liver, biliary tract, gallbladder and pancreas are recognizable structures. During this period, lengthening of the common duct occurs. However, the lumen is occluded by the rapidly proliferating epithelial cells. By the end of the fifth week, recanalization occurs and this start from the proximal portion progressing towards the distal portion of the lumen. By the eighth week, the common duct becomes patent and by 12th week, bile starts to flow from the liver to the second part of the duodenum [5].

The right and the left hepatic ducts converge to form the common hepatic duct just after exiting the liver hilum in 60–70% of the cases [1]. Low convergence of the hepatic ducts is extremely rare and resulted in longer than usual right and left hepatic ducts with resultant shorter than usual common bile duct. The common hepatic duct may or may not be present, depending on the site of insertion of the cystic duct. Frequently, the site of insertion of the cystic duct is anomalous too. Similar to one of our case, the cystic duct may insert to the right hepatic duct. Insertion to the left hepatic duct or low insertion to the more distal portion of the common bile ductal has been reported too [2, 3].

Such anomalies are postulated to be the result of embryological malformations, either due to incomplete recanalization or maldivision of the extrahepatic ductal portion of the embryonic hepatic diverticulum [5, 6].

Clinical presentation of such biliary tree anomalies includes recurrent abdominal pain, jaundice and fever due to biliary stone disease, cholangitis or pancreatitis. Frequently, these anomalies are discovered during the investigations and management of the presenting symptoms. The importance of recognizing such anomalies during surgical procedures such as laparoscopic cholecystectomy, is emphasized as failure to do so may result in iatrogenic biliary tract injury with resultant bile leak and ductal stricture [7, 8].

Both of our cases underwent open cholecystectomy under emergency setting as part of the management of their presenting symptoms. However, intraoperatively the anomaly was missed. This may be due to the presence of inflammatory and oedematous tissues or fibrous
adhesions surrounding the extrahepatic bile ducts making the anomaly less obvious. For the first case, the initial ERCP did not reveal obvious low convergence due to the orientation of the film. This was subsequently detected in the later ERCP.

Biliary tree anomalies may predispose to formation of biliary stones [9]. Both of our patients presented with complications of primary ductal stone. In our cases, we believe such anomalous arrangement may cause abnormal bile flow or bile stasis. We postulated that the long hepatic ducts lie in the edge of the lesser omentum unsupported by solid liver parenchyma and an increase in the intraluminal pressure (i.e., secondary to stone obstruction) will easily stretches the ductal wall and causes ductal dilatation. This perpetuates the vicious cycle of biliary stasis, infection, further stones formation and obstruction.

The presence of recurrent and chronic biliary stones is associated with carcinoma of the biliary tract in the long term [10–11]. Biliary stones may initiate the hyperplasia-carcinoma sequence. As mentioned, it causes bile obstruction and stasis predisposing to infection. These conditions induce chronic inflammation via prolonged exposure of the epithelial cells to bile acids and pancreatic enzymes in the bile. These cause increased cellular proliferation, initiating the multistep progression from epithelial hyperplasia to metaplasia, then dysplasia and finally carcinoma of the biliary tract [10, 12].

Hence, a definitive operative intervention seems prudent despite the lack of consensus in the management of such anomalies, especially in cases with concomitant biliary ectasia and ductal stones. We felt that the decision for definitive management was challenging due to the scarcity of similar cases in literature. We decided to extrapolate the management from the management of adult choledochal cyst. Total excision of the extrahepatic bile duct and gallbladder followed by hepaticoenterostomy is the treatment of choice.

CONCLUSION

Low convergence of hepatic ducts is extremely rare. Such anomaly may be associated with biliary ectasia and ductal stones. Not only it predisposes the patients to recurrent obstruction, infection and pancreatitis, in the long-term it may also lead to carcinoma of the biliary tract. Definitive operative intervention seems prudent, especially in our cases where there are concomitant biliary ectasia and ductal stones.

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Author Contributions

Dayang Azzyati Awang Dahlan – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Nik Azim Nik Abdullah – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Rokayah Julaihi – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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