Anomalous biliary duct mistaken as hilar stricture. A case report

Nachiket Dubale, Anupama N K, Manu Tandon, Pradeep R, DN Reddy, GV Rao

Asian Institute of Gastroenterology

Key words: anomalous extrahepatic biliary anatomy, cholecystohepatic duct, agenesis of CHD, hilar stricture

Abbreviations: CECT, contrast enhanced cat scan; MRCP, magnetic resonance cholangio pancreaticography; PTBD, percutaneous transhepatic biliary drainage; ERCP, endoscopic retrograde cholangio-pancreaticography; RHD, right hepatic duct; LHD, left hepatic duct; CHD, common hepatic duct; CBD, common bile duct; LFT, liver function test; AST, aspartate aminotransferase; ALT, alanine aminotransferase; ALP, alkaline phosphates

Anomalous biliary anatomy is frequently encountered by surgeons during cholecystectomy. Importance of its recognition lies in avoiding serious biliary injuries. One such anomaly is cholecystohepatic duct. We describe rare clinical situation wherein agenesis of CHD along with cholecystohepatic duct was mistaken for hilar stricture.

Introduction

Anomalous drainage of biliary ducts is commonly encountered during cholecystectomies. Knowledge of these variants is essential before performing surgical or non-surgical therapeutic procedures. We describe a case of unique anomalous biliary system with absent common hepatic duct which was mistaken as hilar stricture.

Case profile

49 year old male with no significant past history presented with one week duration of severe epigastric pain abdomen accompanied with jaundice, pruritus and fever. Serum bilirubin was 12 mg/dL (direct 10mg/dL), AST 127 IU/L, ALT 134 IU/L, ALP 300 IU/L (129 upper limit). USG abdomen showed a dilated bilobar intrahepatic biliary radicles, normal common bile duct (CBD) & gallbladder. A diagnosis of extrahepatic biliary obstruction and acute cholangitis was made. Endoscopic retrograde cholangiopancreatogram (ERCP) showed a normal CBD with cut off in its proximal part. Post ERCP, patient developed severe pain and abdominal distension. As luminal perforation was suspected an emergency laparotomy was carried out. However no obvious tear was noted and abdomen was closed. Post surgery, MRCP showed grossly dilated RHD & LHD, CHD was not visualized. Percutaneously the right anterior duct was accessed. As the wire could not be negotiated across the suspected stricture, an external drain was left in situ and he

was referred to our institute with a provisional diagnosis of hilar stricture possibly cholangiocarcinoma.

On admission he was comfortable without any symptoms and a normal LFT. Dilated RHD and LHD with a hilar stricture suggestive of cholangiocarcinoma was seen in a repeat MRCP (**Fig. 1**). CECT abdomen showed bilateral mild IHBD with no portal mass. Cholangiogram from the previously placed external catheter showed dilated RHD and dilated LHD with secondary calculi. LHD could be seen crossing over the midline and draining into a cystic cavity in the region of hilum, along with RHD. CHD was not filled up (**Fig. 2**). ERCP was attempted with the intention to confirm presence of hilar stricture and simultaneously obtain brush cytology. ERCP findings were similar to MRCP with normal CBD and cystic duct and nonopacification of CHD (**Fig. 3**).

Laparotomy was performed in view of endoscopic and radiological failure to access hilar region. Intraoperative cholangiogram identified a cholecystohepatic duct running along the gallbladder wall and opening into the neck region, with agenesis of CHD. CBD was filled up subsequently, opening into duodenum. Hepaticojejunostomy could not be performed due to dense adhesions & deeply seated LHD within liver parenchyma. Hence partial cholecystectomy with cholecystodeudonostomy was done. Percutaneous cholangiogram on 8th postoperative day from the Rt PTBD catheter showed dilated RHD, LHD with free flow of contrast into duodenum via cholecystoduodenostomy. PTBD drain was removed before discharge.

Discussion

The biliary tract is notorious for its variable anatomy. The classical textbook anatomy of the biliary tract is seen in less than 50% of cases. This variable anatomy adds to the operative difficulties, especially in the presence of significant changes occurring as a

*Correspondence to: Nachiket Dubale; Email: drnachik@yahoo.co.in
Submitted: Jul/07/2010; Revised: Jul/11/2010; Accepted: Jul/16/2010
Previously published online: www.landesbioscience.com/journals/jig
DOI: 10.4161/jig.1.1.14600

Figure 1. MRCP showing dilated RHD, LHD with non-projected CHD, normal CBD and PD. PTBD catheter in situ.

Figure 2. Cholangiogram via PTBD showing dilated RHD communicating with LHD & nonopacification of common hepatic duct.

Figure 3. ERCP showing CBD, Gallbladder with cystic duct and cholecystohepatic duct (arrow).

Figure 4. Different types of cholecystohepatic duct.

result of the disease process itself.¹ Anomalies of the extrahepatic biliary tree may arise as a result of aberrations of the normal embryological development. One such rare anomaly is the hepatico-cystic duct, wherein the RHD & LHD drain directly into the gallbladder, with agenesis of the CHD & CBD, also been referred to as cholecystohepatic duct, congenital absence of the common bile duct, transverse lie of the gallbladder, or, gallbladder interposition by various authors.^{2,3} Cholecystohepatic duct is the result of the persistent connection between fetal hepatic duct and the gall bladder with failure of recanalisation

of the CHD resulting in atresia.^{4,5} Incidence was found to be less than 0.85% in a large series of cholecystectomies. Several studies have also shown a combined incidence of cystohepatic and cholecystohepatic ducts between 0.2 to 0.3%.⁶

Various patterns of hepaticocystic ducts have been recognized (**Fig. 4**).

Type I refers to the absence of the common hepatic duct and the right and left ducts drain separately into the gallbladder; Type II is said to be present when the right and left hepatic ducts unite upon entering the gallbladder; Type III refers to a common

hepatic duct that enters the gallbladder, and in Type IV multiple small bile ducts connect the intrahepatic biliary system with the gallbladder. Type III is further subdivided into the common hepatic duct entering the superior wall of the gallbladder (III A), neck (III B), posterior gallbladder wall (III C), and, the fundus (III D). Our patient probably belongs to the type III B.⁷

In our case patient, in view of short history and normal looking porta on CECT, congenital biliary duct anomaly was thought of. Surgical intervention was decided in view of failure of percutaneous and endoscopic methods to access and opacify CHD 'stricture'. The intention was to delineate biliary tree and

rule out cholangio-carcinoma. As reported in the previous case reports, pre-operative imaging in our case too, failed to diagnose this anomaly probably due to the small calibre of the ducts.⁸

In the best of our knowledge symptomatic cholecysto-hepatic ducts have not been reported so far.

Disclosure

Authors have no conflicts of interest financial or otherwise.

References

1. Sharma V, Saraswat VA, Baijal SS, Choudhuri G. Anatomic variations in intrahepatic bile ducts in a north Indian population. *J Gastroenterol Hepatol* 2008; 23:e58-62.
2. Walia HS, Abraham TK, Baraka A. Gall-bladder interposition: a rare anomaly of the extrahepatic ducts. *Int Surg* 1986; 71:117-21.
3. Lamah M, Karanjia ND, Dickson GH. Anatomical variations of the extrahepatic biliary tree: review of the world literature. *Clin Anat* 2001;14:167-72.
4. Kihne MJ, Schenken JR, Moor BJ, Karrer FW. Persistent cholecystohepatic ducts. *Arch Surg* 1980;115:972-4.
5. Moore KL, Persaud TVN. The developing human. 8th edition. Philadelphia, PA: W.B. Saunders; 2008. 219-223.
6. Champetier J, Létoublon C, Alnaasan I, Charvin B. The cystohepatic ducts: surgical implications. *Surg Radiol Anat* 1991; 13:203-11.
7. Losanoff JE, Jones JW, Richman BW, Rangnekar NJ. Hepaticocystic duct: a rare anomaly of the extrahepatic biliary system. *Clin Anat* 2002; 15:314-5.
8. Schofield A, Hankins J, Sutherland F. A case of cholecystohepatic duct with atrophic common hepatic duct. *HPB (Oxford)* 2003; 5:261-3.