

Gallbladder duplication in a child with choledochal cyst: a rare surgical surprise

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DESCRIPTION

A 6-year-old boy presented with epigastric pain and intermittent jaundice for 2 months. There was no history of fever, weight loss or anorexia. General physical examination did not reveal any abnormality. Abdomen was soft, non-tender and without any lump or organomegaly. Laboratory test showed mildly elevated (183 U/L) alkaline phosphatase level whereas other parameters were within normal range.

Abdominal ultrasound showed bilobar intrahepatic biliary radicle dilation with fusiform dilation of extrahepatic bile duct. Gallbladder was distended with normal wall thickness. Multiple tiny stones were noted in gallbladder and common bile duct lumen. Magnetic resonance cholangiopancreatogram also revealed Todani type 1 choledochal cyst (CDC) with cholelithiasis. The gallbladder was reported to be mildly distended and filled with multiple tiny calculi.

The patient was planned for CDC excision with cholecystectomy and Roux-en-Y hepaticojejunostomy. Intraoperatively, two bulges were found in the gallbladder fundal region underneath the peritoneal covering (figure 1). On further dissection, two gallbladders with separate cystic ducts were found (figure 2A,B). CDC measured approximately 2.5 cm. Multiple pigmented (black) stones were present in gallbladder and CDC lumen. Complete excision of the CDC along with cholecystectomy was performed in our patient. The cyst was transected proximally at the common hepatic duct, just below the level of hilum. The tapering of the bile duct was identified distally within the head of pancreas and transection was done at that level. The distal bile duct stump was ligated carefully to avoid postoperative pancreatic fistula. Both gallbladder lobes were excised. Biliary-enteric flow is re-established through a wide mucosa-to-mucosa Roux-en-Y hepaticojejunostomy at the level of the hilum. Postoperative recovery was uneventful and the patient was discharged on fifth day following surgery.

Gallbladder duplication is a rare congenital anomaly with incidence of 1 in 4000 patients.¹ The incidence is twice more common in women than in men.² CDC is another rare anomaly of the biliary tract characterised by abnormal dilation of intrahepatic and/or extrahepatic bile ducts. The association of duplex gallbladder and CDC is extremely rare and only two such case have been reported in the medical literature.^{3,4} One of the reported case was based only on imaging findings as the patient was asymptomatic and did not undergo surgery.⁴

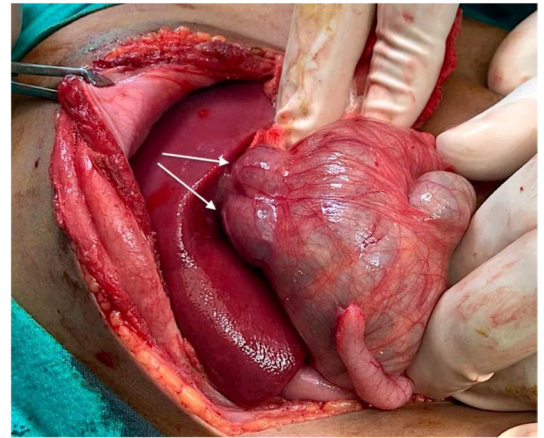


Figure 1 Double gallbladder (arrows) under peritoneal covering seen on abdominal exploration.

Gallbladder duplication is classified according to Boyden's classification. Type-I anomaly refers to a split or bilobed gallbladder whereas type-II anomaly comprises of two separate gallbladders, each with their own cystic duct.⁵ Differential diagnosis of a duplex gallbladder includes gallbladder diverticula, gallbladder fold, Phrygian cap and focal adenomyomatosis. Double gallbladder as such is not an indication for prophylactic cholecystectomy. However, when the gallbladder becomes diseased and symptomatic, simultaneous removal of both gallbladders is indicated, even if one of the lobes is normal. This is done to avoid repeat cholecystectomy in case of recurrent cholelithiasis and cholecystitis.⁶ Gallbladder duplication may occasionally be misinterpreted as CDC of diverticular variety, that is, Todani type II. Also, when double cystic duct is encountered during Calot's triangle dissection, possibility of an aberrant right posterior

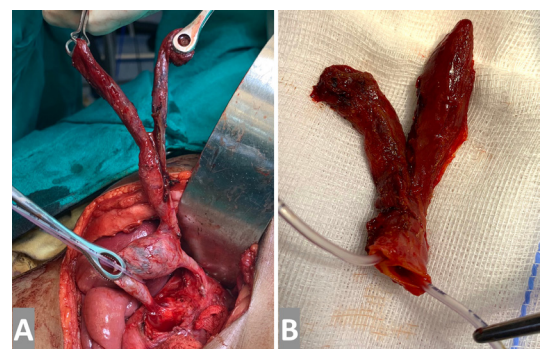


Figure 2 (A) double gallbladder with dissected choledochal cyst; (B) gallbladders with separate cystic ducts as seen on cut section.



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Images in...

sectoral duct should always be ruled out. Furthermore, cystic artery should be carefully delineated before dividing, as anomalous right hepatic artery may also be associated with double gallbladder. Intrahepatic location of one of the gallbladder lobes may produce difficulty in its identification and dissection intraoperatively.^{3 6 7} In our patient, gallbladder duplication was not associated with anomalous hepatic artery or aberrant sectoral hepatic duct. In our patient, double gallbladder was detected intraoperatively while performing CDC excision.

CDC has been classified into five types using the Todani classification system. Type 1 is the most common variant and account for 85%–90% of all cases. Although majority of the CDC can be classified according to Todani classification, few rare variants have been described that do not fit into this classification system. These include variants such as cystic dilatation of the cystic duct, porta hepatis or isolated involvement of the right or left hepatic duct.⁸ Our patient had type 1 CDC with fusiform dilation of extrahepatic biliary tree. Since CDC is associated with high risk of complications such as cholangitis, pancreatitis and malignant transformation, cyst excision is indicated once the diagnosis is established.

In our patient, the diagnosis of duplex gallbladder was missed on preoperative imaging and was discovered as a surgical surprise. Because CDC is associated with biliary anomalies, a

high index of suspicion is required for diagnosing gallbladder duplication in these cases. It remains unclear whether the coexistence of duplex gallbladder and CDC is just coincidental or has some etiological correlation.

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Learning points

- ▶ The present case describes an unusual association of two rare congenital conditions.
- ▶ Choledochal cyst and symptomatic duplex gallbladder are indication for surgery
- ▶ Knowledge of anatomy and high level of suspicion for associated biliary anomalies in a case of choledochal cyst is key for safe surgery.

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