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Case Report

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Type VI B choledochal cyst: a rare entity

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ABSTRACT

Choledochal cysts (CDC) are congenital dilatations of extra hepatic biliary system with or without involving intrahepatic bile ducts. Traditionally classified into five types with emerging type VI, involving dilatation of cystic duct, which is the rarest. It is seen in middle-aged females. Most are symptomatic with common symptom being abdominal pain. Magnetic resonance cholangiopancreatography (MRCP) is diagnostic. Surgical treatments range from cholecystectomy to Roux en Y hepaticojejunostomy. They are at a risk of malignant transformation. Hence should be looked for when encountered with congenital biliary system anomalies. We presented a case of a young male with type VI B CDC and its management. A brief case report with review of literature is presented.

Keywords: Choledochal cysts, Type 6 choledochal cysts, Congenital biliary anomalies

INTRODUCTION

CDC are congenital or acquired cystic dilatations of any portion of the intrahepatic and/or extrahepatic bile ducts, most commonly involving common bile duct (CBD).[1-3] Traditionally CDC have been classified into 5 types.[4] Here we present rare case of combined dilatation of cystic duct and CBD reported as Variant of type VI known as type VI B, classified by some as type ID as addition to Todanis type I CDC along with it's surgical management.[1,2]

CASE REPORT

A 26-year-old male patient presented with a complaint of intermittent right upper abdominal pain aggravated by fat rich meals for one year. Clinical examination and laboratory tests were normal. Ultrasound revealed outpouching of proximal CBD of size 3.8×2.6 cm. There was no intrahepatic biliary radicle dilatation (IHBRD). MRCP revealed a fusiform dilatation of proximal two third of CBD, 1.9 cm in diameter and 4.2 cm in length with a dilated cystic duct with wide opening of cystic duct in common hepatic duct (CHD) (Figure 1).

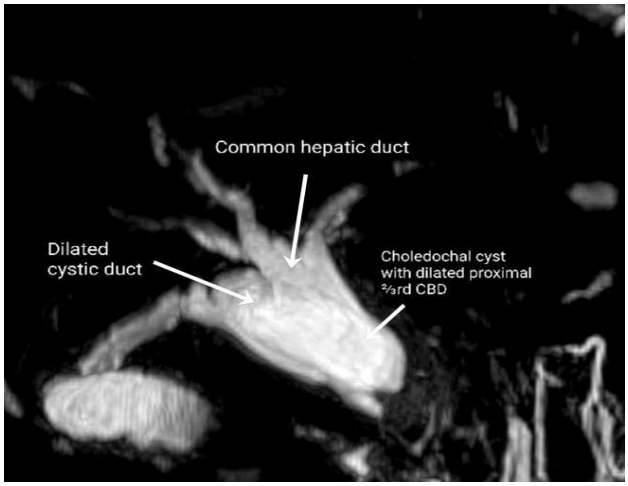

Figure 1: MRCP image showing dilated cystic duct and proximal CBD.

Mild prominence of right and left hepatic ducts and normal distal most part of CBD with diameter of 2.8 mm. Main pancreatic duct diameter in body was 2 mm with no evidence of abnormal pancreaticobiliary duct junction (APBDJ) and no IHBRD. Due to the wide opening of the cystic duct in CHD and wide proximal CBD, a decision of complete excision of CDC and gall bladder along with hepaticojejunostomy was taken. Intraoperatively gallbladder (GB) and cyst were carefully dissected, cholecystectomy with excision of dilated CBD was done (Figure 2).

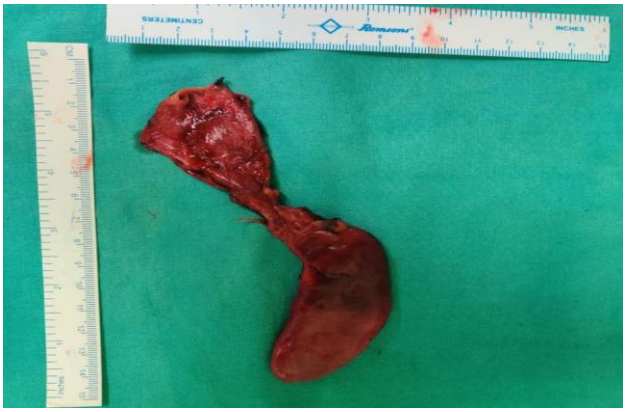

Figure 2: Surgical specimen showing dilated cystic duct and gall bladder.

Lower end of CBD was closed and Roux-en-Y hepaticojejunostomy was performed. Subhepatic and pelvic drains were placed which were removed subsequently. The postoperative course was uneventful. Histopathology specimen revealed a wide opening of cystic duct into CBD with proximally dilated CBD. GB microscopy revealed features of chronic cholecystitis. Follow up of 1 year has shown patient to be symptom and disease free.

DISCUSSION

Alonso-Lej et al in 1959 initially described three types of CDC.[5] These were later modified by Todani et al to include a total of five variants.[4] The sixth variant consisting of isolated cystic duct dilatation, was described in the recent past. Very first case of which was described by Bode et al in 1983.[6]

Serradel et al modified the classification of Todani to include isolated cystic duct cysts and proposed novel type VI CDCs.[7] Yoon in 2011 described the imaging features of choledochal cysts with involvement of the cystic duct in three cases; in one of the cases only cystic duct was dilated and in other two both the cystic and common bile ducts were involved. Then he proposed that these variant anomalies be included in the classification of choledochal cysts.[8] Michaelides et al in a series of six cases have proposed the classification of this type of choledochal cyst as a new subtype of Todani I cyst, namely type ID.[1]

Bhoil et al proposed that type VI cysts can be further divided into two types: type VI A as isolated dilatation of the cystic duct and type VI B involving combined dilatation of cystic duct as well as the common bile duct (CBD) (Table 1).[3] This classification guides the definitive management. Sugarman et al reported 2 cases which were reported in MRCP as type Ia and type II CDC but intraoperative findings revealed each of type VI A and B CDC which changed the course of management.[2] This emphasizes the importance of knowledge of these newer types and the need for the further extension of the traditional classification.

Table 1: Studies published describing type VI B choledochal cyst.

Study	Number of cases	Features of CDC	Classification given
Yoon[8]	3-with involvement of cystic duct	2 had involvement of both the cystic and CBD	Not given
Michaelides et al[1]	6	Dilatation of the CHD, CBD and dilatation of the central portion of the cystic duct	Type 1D
Bhoil et al[3]	1	Combined dilatation of cystic duct as well as the CBD	Type VI B
Sugarman et al[2]	2	1 isolated cystic duct dilatation, 1 combined cystic and CBD dilatation	Type VI A and B respectively
Present study	1	Combined dilatation of cystic duct as well as the CBD	Type VI B

Most of the cystic duct lesions are symptomatic, common symptom being epigastric and/or right upper abdominal pain aggravated by a fatty meal as was in the present case. Though exact etiology of these cysts is unknown, an APBDJ causes ectasia of CBD and cystic duct.[6,8-11] Not all cases have APBDJ causing CDC, in Hirschsprung's disease, a focal aganglionosis of the cystic duct is said to be the cause.[1,10,12] The initial investigation is commonly abdominal ultrasonography but the ideal is MRCP to delineate the entire biliary system anatomy including the IHBRD, GB wall thickening, gallstones, cystic duct course, CBD involvement, and detection of APBDJ. Endoscopic retrograde cholangiopancreatography (ERCP) is invasive and provides the same information regarding the biliary system as MRCP. ERCP and Tc-99m hydroxy

imino diacetic acid (HIDA) scan can diagnose but are not commonly used.[13,14]

Management of type VI A is simple cholecystectomy, if the opening of the cystic duct into CBD is narrow. Which can be done laparoscopically by clipping the cyst opening into the CHD.[12,15,16] If there's a wide opening, cyst and CBD excision along with Roux-en-Y hepaticojejunostomy is required.[2,4,8,9,12,15,16] Rationale for complete surgical excision of the cyst along with cholecystectomy is that the epithelium of these cysts is at risk of developing biliary intraepithelial neoplasia (BIN). Multiple cut sections of the histopathology specimen must be analyzed for the same.[12,15,17]

Very few cases of type VI B choledochal cysts have been reported in the literature until now, summarized in Table 1.

CONCLUSION

Knowledge of this new variant of choledochal cyst is essential for early diagnosis and treatment. Surgeons may find type VI CDC incidentally intraoperatively while operating on other benign hepatobiliary conditions and prior knowledge is required for better management. With increasing availability of MRCP for diagnosing hepato-biliary-pancreatic pathologies knowledge of new biliary cystic dilatation is the need of the hour.

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