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Diagnostic Dilemma Between Todani Type II Choledochal Cyst and Duplication of Gallblader in an Adult Patient

Lorentziadis ML[1], Amir Mounir Hussein[2], Moustafa Mahmoud Nafady Hego[2], Ghada M Almakhaita[3]

Abstract

Congenital cystic abnormalities of the biliary tree are rare and sometimes it can be difficult to differentiate preoperatively whether the anomaly is a duplication of the gallbladder or a choledochal cyst.We present a case of a young female patient who was investigated for atypical chronic symptoms. Imaging studies with u/s and MRCP confirmed the presence of a cystic lesion of 5x6cm next to the gallbladder which could not be differentiated from choledochal cyst type II or duplication of the gallbladder. It was excised laparoscopically, with concomitant cholecystectomy. On histology, proved to be a choledochal cyst type II. Laparoscopic approach in such cases is feasible and should be undertaken cautiously applying the technical tips necessary for patient's safety.

Key words: *Choledochal cyst; duplication of gallbladder; laparoscopic excision; biliary tract anomaly*

Introduction

Choledochal cysts are rare (1:13000-1:2000000 child births) [1,2] congenital dilatations or diverticulum of all or part of the biliary tree occurring more frequently in females (M/F ratio: 1/3-1/8) [3]. They are classified in five subtypes. In the imaging studies, choledochal cyst type II can mimic duplication of the gallbladder [3,4]. In this paper, we present a case in where preoperatively the patient was diagnosed with a cystic anomaly of the extrahepatic biliary tree but the imaging studies were inconclusive whether it was a choledochal cyst or gallbladder duplication.

Case Presentation

A 25-year-old female patient appeared at our department complaining of chronic vague right upper abdominal pain, mainly after meals. She had no medical or surgical history. Physical examination and laboratory tests were normal. In the abdominal ultrasound, a cystic lesion 5x6cm next to the gallbladder was evident containing sludge, without dilatation of the extrahepatic biliary tree. The gallbladder was partially distended with no feature of cholelithiasis. MRCP confirmed the presence of the cyst which was inseparable from the gallbladder, in direct contact with the right branch of the portal vein (Figure 1).

The patient underwent laparoscopic resection of the cystic lesion and the gallbladder (Figure 2). The intraoperative cholangiogram, which is obligatory in such cases, showed that there was no communication between gallbladder and the cyst (Figure 3). The resection was uneventful except from some bleeding at the site of contact with the right portal branch which was successfully

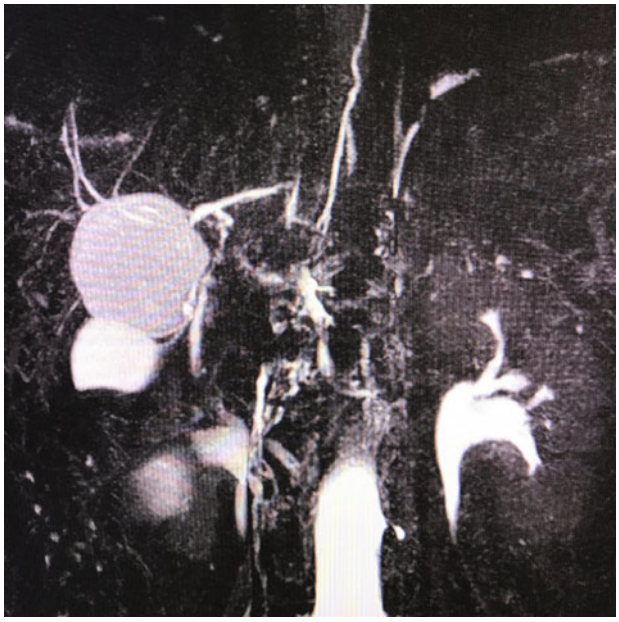

Figure 1. MRCP showing the cystic lesion in close proximity with the gallbladder. A such an image could represent choledochal cyst or duplication of the gallbladder.

[1]*Consultant, General Surgery*

[2]*Associate Consultant, General Surgery*

[3]*Consultant Radiologist*

Imam Abdulrahman bin Faisal Hospital, Dammam, KSA

Corresponding author: Michail L. Lorentziadis
Tel.: +30 6932 478646, e-mail: lorentziadism@gmail.com

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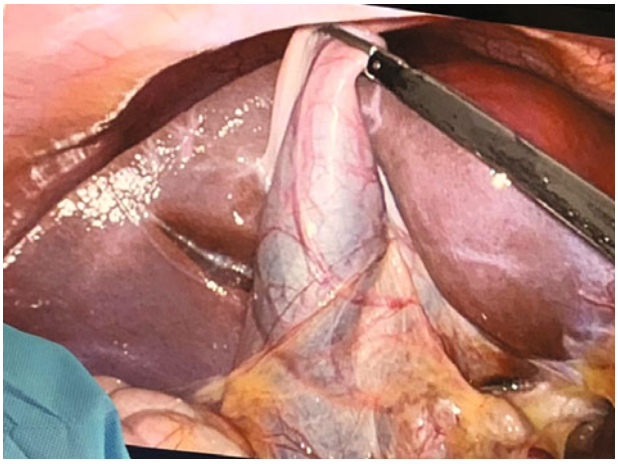

Figure 2. Operative picture of the cystic lesion behind and in close contact with the gallbladder.

controlled (Figure 4). Histologic examination showed that the lesion was a type II choledochal cyst. Eight months later, the patient remains well and symptom free with unremarkable abdomen ultrasound.

Discussion

The aetiology of choledochal cysts is unknown .There are various hypotheses for the aetiology of this rare congenital abnormality of the biliary tree. Based on the observation that in most cases (57-96%) an anomalous pancreaticobiliary confluence is present, it is postulated that this anomaly affects the function of the sphincter of Oddi, causing pancreaticobiliary reflux, which results in inflammation and ectasia leading to cystic degeneration of the duct wall. Other hypotheses, include obstruction of the CBD, spasm of sphincter of Oddi and abnormal ganglion cells of the CBD. All theories conclude that the anatomical or functional obstruction in the distal CBD increases the intraductal biliary pressure and causes the development of choledochal cysts [6, 7].

The most commonly used classification is Todani's, who modified the 1959 Alonso-Lej classification [6,8]. He proposed five types of choledochal cysts as follow: Type I, cystic (A) or focal (B) or fusiform (C) dilatation of CBD (50-80%). Type II, saccular diverticulum of the CBD (2%). Type III cystic dilatation of the intramural part of the CBD-choledochocele (1,4-4,5%).Type IV, involving the intra and extrahepatic part(IVa) or only the extrahepatic part (IVb) of the biliary ducts. Type V, multiple segmental intrahepatic bile ducts – Caroli disease (20%).

Our patient appeared with nonspecific upper abdominal pain and this is the case in most patients, since the classical triad jaundice, right upper quadrant mass and

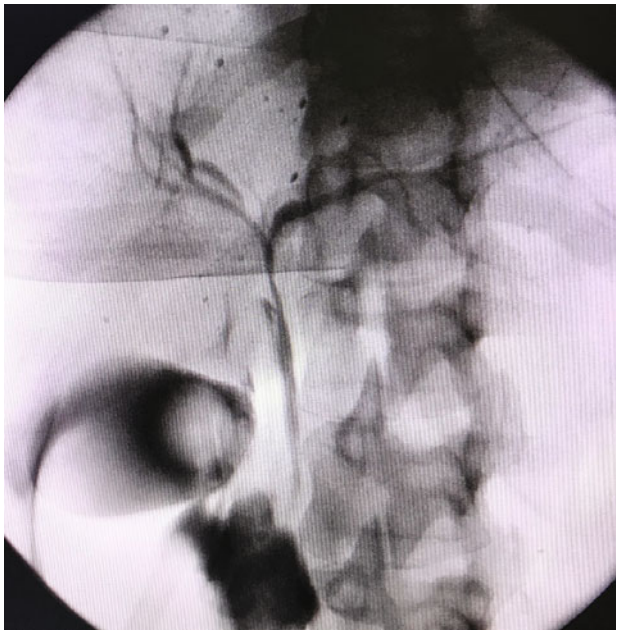

Figure 3. Intraoperative cholangiogram which shows the lack of communication between the cystic lesion and the gallbladder.

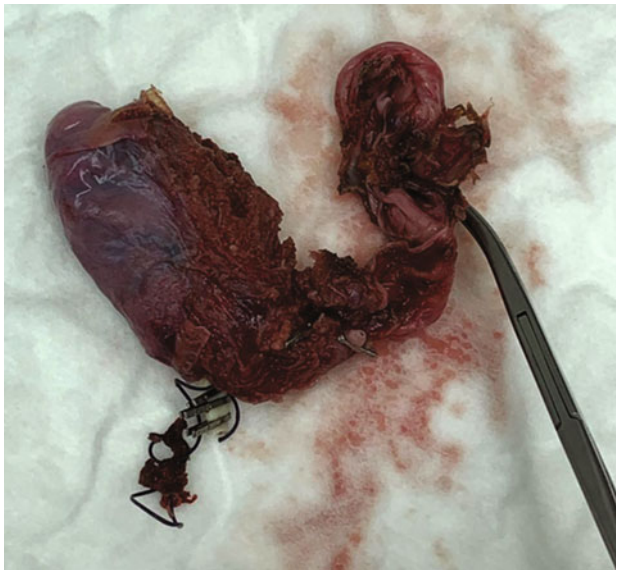

Figure 4. Excised specimen of the choledochal cyst and the gallbladder.

abdominal pain is only evident in 0-17% of cases [9]. In adults, choledochal cysts may be an incidental finding in imaging investigations. They can be complicated by lithiasis, pancreatitis, cholangitis, biliary cirrhosis, portal hypertension secondary either to portal vein thrombosis or Caroli's disease and malignancy [6, 7].The aetiology of malignant change is unknown and it is postulated that the stagnation of bile which produces bile carcinogens leads to glandular

hyperplasia and epithelial malignancy. Todani type II and III choledochal cysts rarely undergo malignant changes [10].

The differential diagnosis include gallbladder diverticulum or duplication, especially in choledochal cysts Type II, located in the upper (58%) or middle (21%) third of the CBD. Histopathology, as in our case, confirms the final diagnosis since the choledochal cyst lacks muscular wall with epithelial lining. Ultrasound is the front line imaging method for investigating the biliary tree pathology but MRCP is the gold standard for diagnosis [7, 8].

Incidental finding of a choledochal cyst is an indication for surgery in order to prevent the development of complications .On the contrary, asymptomatic or incidentally discovered duplication of gallbladder is not an indication for surgery [10]. The operative strategy depends on the type of the cyst and the hepatobiliary pathology. In principle, the choledochal cyst with the draining duct should be excised and reestablish the continuity of bile flow. Resection of a Type II choledochal cyst can be done laparoscopically or by laparotomy.The intraoperative cholangiogram is compulsory since it clarifies the anatomical relations of the cyst with the CBD and especially the way they communicate. If they are connected by a duct draining to the CBD, as in our case, the choledochal cyst is resected like a gallbladder with clip ligation of the draining duct. If the connection is larger, the defect of the CBD wall can be closed primarily or with a T-tube. If the cyst arises from the intra-pancreatic portion of the CBD it is easier to drain it into the duodenum [1, 8].

Conclusion

Type II choledochal cysts are rare and in case they drain into the biliary tree, it may be difficult to differentiate from duplication of the gallbladder. In such cases, the intraoperative cholangiogram and meticulous laparoscopic resection is the recommended treatment. Histology confirms the final diagnosis since choledochal cysts lack muscular wall with epithelial lining.

Conflict of Interest: *The authors declare that they have no conflict of interest.*

Ethical Approval – Informed Consent: *The authors declare that the study has been approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki. Also all patients gave their written informed consent prior to their inclusion to the study.*

References

1. Khandelwal C, Amand U, Kumar B, et al. Diagnosis and management of choledochal cysts. Indian J Surg 2012;74:29-39.
2. Soreide K, Korner H, Havnen J, et al. Bile duct cysts in adults. Br J Surg 2004;91:1538-48.
3. Kyung Lee H, Park Jin S, Yi Ha B, et al. Imaging features of adult choledochal cysts: A pictorial review. Korean J Radiol 2009;10:71-80.
4. Gorecki PJ, Andrei VE, Musacchio T, et al. Double gallbladder originating from left hepatic duct: A case report and review of the literature. J SLC 1998;2:337-8.
5. Quaissi M, Kianmanesh R, Belghiti J, et al. Todani type II congenital bile duct cyst. European multicenter study of the French Surgical Association and literature review. Ann Surg 2015;262:130-8.
6. Mesleh M, Deziel D. Bile duct cysts Surg Clin N Am 2008; 88:1369-84.
7. Metcalfe M, Wemyss-Holden S, Maddern G. Management dilemmas with choledochal cysts. Arch Surg 2003;138:333-9.
8. Lipsett PA, Pitt HA. Surgical treatment of choledochal cysts. J Hepatobiliary Pancreat Surg 2003;10;352-9.
9. Sherman P, Kolster E, Davis C. Choledochal cysts; Heterogenicity of clinical presentation. J.Pediatr.Gastroenterol. Nut. 1986; 5:867-72.
10. Sastry AV, Abbadessa B, Wayne MG, et al. What is the incidence of biliary carcinoma in choledochal cysts, when do they develop, and how should it affect management? World J Surg 2015;39:487-92.