International Surgery Journal
Bhasin SK et al. Int Surg J. 2014 Aug;1(2):112-115
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902
DOI: 10.5455/2349-2902.isj20140813

Case Report

Missed choledochal cyst: a rare presentation and review of literature

Sanjay Kumar Bhasin*, Tariq Parvez Azad, Nasib C. Digra, Mubashar, Gopal Sharma

Department of Surgery, Govt. Medical College, Jammu, J & K, India

Received: 24 July 2014
Accepted: 5 August 2014

***Correspondence:**
Dr. Sanjay Kumar Bhasin,
E-mail: sanjaykbhasin@yahoo.co.in

ABSTRACT

Choledochal cysts remained uncommon and less reported anomalies of biliary system till recently. "Forme Frustae choledochal cyst" was coined by Lilly in 1985 and the entity can very easily be missed during first surgery. The present case is a 35 years old female who was admitted for elective Cholecystectomy with Choledochotomy. Due to excessive T-tube drainage for prolonged period of time and fusiform dilatation of CBD on postoperative T-tube cholangiogram and MRCP, patient was subjected to second surgery and hepaticojejunostomy was done successfully and patient discharge from hospital on 9th postoperative day. The case is being presented due to it's rarity vis a vis reviewing the literature on the subject.

Keywords: Choledochal cyst, Choledocholithiasis, Bilioenteric anastomosis, T-tube

INTRODUCTION

Choledochal cyst was reported first time in 1852[1] but it was Vater[2] in 1720 who first time described congenital dilatation of the extra hepatic Biliary ducts.

Choledochal cysts remained uncommon and less reported anomalies of biliary system as there were only 955 reported cases of choledochal cyst in the literature till 1975.[3]

Alonso-Lej et al. (1959)[4] initially gave classification of choledochal cysts and Todani et al. (1977) further went on expanding the classification into 05 types and included concept of APBJ that has been accepted worldwide.[5]

In 1985 Lilly et al. coined the term 'Forme Frustae choledochal cyst' with characteristic features of choledochal cyst except for cystic component. However first report of such type was published by Okada et al. in 1981 when it was called as 'common channel syndrome'. Now this is a variant of choledochal cyst that has minimal or no dilatation of the extra hepatic bile duct.[6-8]

The present case under report is a case of "Forme Frusta choledochal cyst" that was missed during first surgery (Cholecystectomy with choledochotomy with T-tube placement). In view of rarity of the case we present it here.

CASE REPORT

Thirty five years old, married female, Hindu by religion, belonging to low socioeconomic strata was admitted as a case of ultrasound proved symptomatic cholelithiasis with solitary stone in the CBD. There was no history of jaundice or any other co-morbid condition. The ultrasound report revealed multiple stones in the Gall bladder and 15 mm stone in the distal end of CBD, with 13 mm CBD diameter. In view of low economic status of the patient she could not afford ERCP retrieval of the CBD stone. So after routine investigations, she was subjected to elective Cholecystectomy and choledochotomy with T-tube placement in the CBD after retrieval of the stone from CBD. Intra-operative findings were that of well distended gall bladder without any adhesions and normal caliber cystic duct. The CBD was

dilated (18-20 mm) and a solitary stone pigment stone of about 12-13 mm was retrieved from it. The proximal and distal patency of the CBD could be ensured with 7F infant feeding tube. Choledochotomy was sutured over T-tube with 3-0 vicryl. Post-operatively the T-tube drainage ranged from 600 ml to 2 letters till 9th post-operative day when we did a T-tube Cholangiogram that showed saccular dilatation of the CBD with normal intra-hepatic biliary channels and no flow of dye in the duodenum (Figure 1).

On 12-13th post-operative day there was no bile drainage from T-tube. We clamped the T-tube for 04 days till 18th post-operative day. The patient experienced two episodes of mild pain abdomen during T-tube clamping, however, there was no fever and no evidence of jaundice either clinically or biochemically during this period. The Ultrasonography done on 17th post-operative day revealed post-Cholecystectomy status of GB with 15 mm CBD and no intra-peritoneal collection. T-tube was again opened on 19th post-operative day for another 07 days when the T-tube drainage ranged from 300 ml to 1.9 lt. We subjected the patient to MRCP; which showed dilatation of CBD with partial distal patency (Figure 2).

The T-tube was kept opened thereafter till 45th postoperative day when again T-tube cholangiogram was performed that revealed partial flow of dye into the duodenum and the dilatation of CBD persisted with normal intra-hepatic biliary channels in the second cholangiogram also (Figure 3).

With provisional diagnosis of missed choledochal cyst we re-explored the patient and performed cystectomy with Roux-en-Y hepaticojejunostomy on 53rd day of the first surgery. The cyst (Figure 4) was subjected to histopathology examination that revealed findings suggestive of choledochal cyst. Patient was discharged on 9th post-operative day after second surgery. Patient has remained in follow up for 12 weeks without any specific complains.

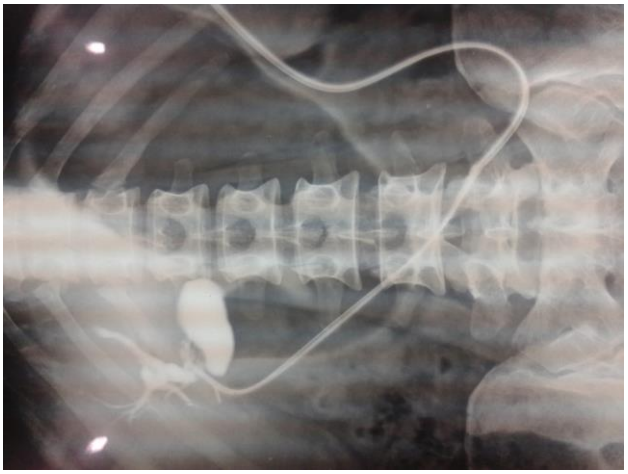

Figure 1: T-tube cholangiogram on 10th post-operative day showing dilatation of CBD.

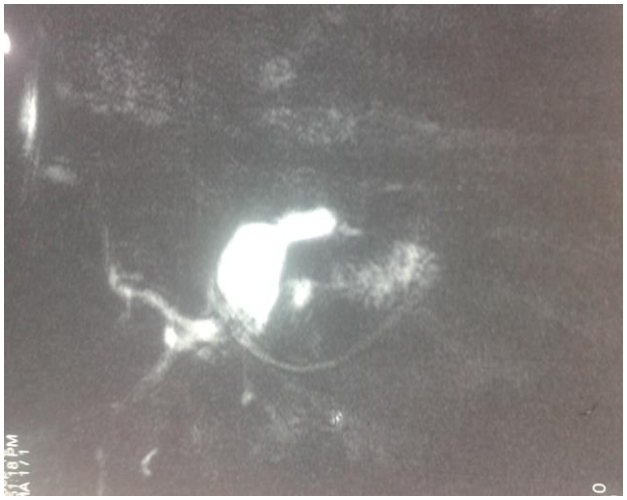

Figure 2: MRCP of the patient on 23rd post-operative day again showing dilatation of CBD.

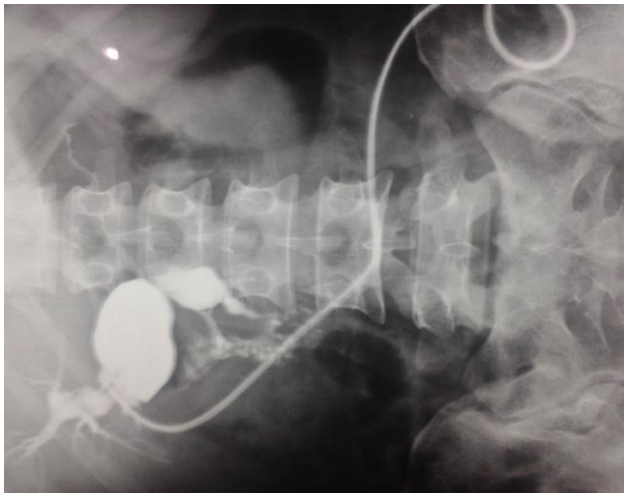

Figure 3: T-tube cholangiogram before second surgery showing dilatation of CBD.

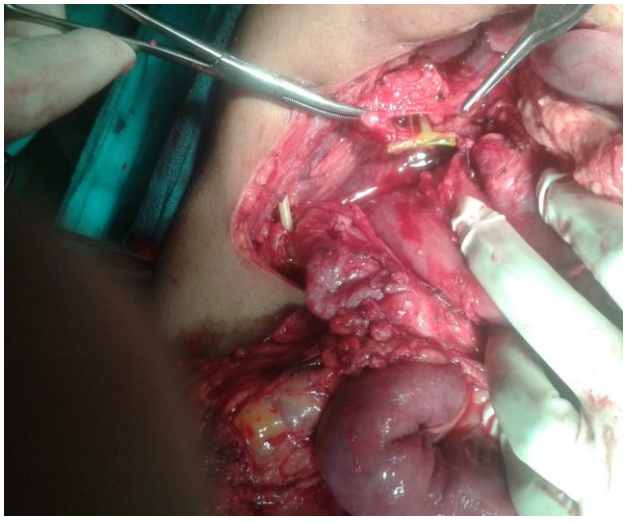

Figure 4: T-tube seen in the cystic dilatation of the CBD during second surgery.

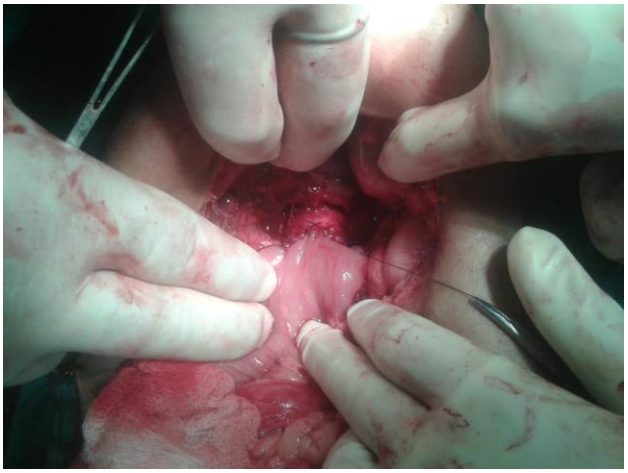

Figure 5: Hepaticojejunostomy completed after complete cyst excision.

DISCUSSION

Bile duct cysts or biliary cysts commonly referred to as choledochal cysts are congenital cystic dilatations of the biliary tree, but in nearly 20% of the patients the diagnosis is delayed until adulthood.[3]

The etiology of choledochal cysts remains speculative. The most widely accepted hypothesis is the presence of an anomalous pancreatico-biliary ductal confluence (APBDJ) proximal to the regulatory control of the sphincter mechanism within the duodenal wall. The observation that biliary cysts are more common in the extra hepatic biliary tree supports this hypothesis.[8,9]

Ultrasonography as well as ERCP provide adequate information about the intra- and extra-hepatic biliary tree and are extremely useful investigations. In addition ERCP defines the anatomy of the biliary tract accurately and reveals the presence of any associated intraductal pathology or an APBDJ. MRCP, being a non-invasive procedure, is emerging as a favored alternative to ERCP, but it has a lower accuracy in detection of APBDJ.[10-12]

There was a 61.4% incidence of calculi within the choledochal cyst and in 57.1% of them the gall bladder also contained calculi. Of the seven patients who had hepatolithiasis, six belonged to type IVa cysts. The seventh patient had type I cyst with a stricture at the distal end of the right anterior sectoral duct. The dilated proximal portion of the duct contained calculi.[13,14]

Ours was a young female who first underwent cholecystectomy with choledochotomy with T-tube placement, followed by Roux-en-Y hepaticojejunostomy for post-operative diagnosis of type-I choledochal cyst, as there was excessive and prolonged T-tube bile drainage. Banerjee Jesudason[15] reported that fourteen patients had undergone cholecystectomy elsewhere and 10 of them had also undergone common bile duct exploration. All these patients had been erroneously diagnosed as calculous cholecystitis, with or without associated choledocholithiasis. In all the missed cases of choledochal cyst second exploration and standard surgical protocol was followed.

Uta Waidner et al. suggested that type I cyst should be considered in the differential diagnosis of any patient with ductal dilatation. In their case report a 19 years old female underwent laparoscopic fenestration of hepatic cyst, with abdominal drainage. Pt developed post-operative jaundice and biliary fistula. Post-operative ERCP failed to detect biliary system and dye leaked into the abdominal cavity. The CECT review in referral hospital revealed type I choledochal cyst that was managed by Cholecystectomy with Cystectomy with Roux-en-Y hepaticojejunostomy.[16] There is another case report by Fransisca J. Siahaya et al. of uncommon mixed type I and II choledochal cyst that was not taken care of during laparoscopic Cholecystectomy and subsequently needed second surgery after 02 months.[17]

In the present case report, with the initial diagnosis of cholelithiasis with choledocholithiasis we performed Cholecystectomy with choledochotomy and retrieval of CBD stone followed by closure of choledochotomy over a T-tube. The excessive T-tube drainage compelled us to revisit and further investigate the patient that revealed a Type I choledochal cyst. After searching the pubmed and the surgical literature available on the subject we hardly find any such presentation or case report. Hence in view of rarity of the case it is been reported.

CONCLUSION

Choledochal cysts are rare in children and yet further rare in adult population. The cases of missed choledochal cysts during the first surgery and re-exploration thereafter has been sporadically reported in the literature, but diagnosing the missed choledochal cyst in the manner discussed in the article is rare and probably reported for the first time. When pre-operative ERCP in CBD stones has not been done and intraoperative findings are consistent with moderately dilated CBD with normal looking cystic duct and gall bladder one must keep in mind the possibility of type I choledochal cyst or “Forme Frustae choledochal cyst”.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Vater A. Dissertation inauguralis medica poes diss qua scirrhis viscerum dissert, c.s.ezierus. Edinburgh Univ Library. 1723;70:19.
2. Douglas AH. Case of dilatation of the common bile duct. Month J Med Sci. 1852;14:97-101.
3. Flanigan DP. Biliary cysts. Ann Surg. 1975;182;635.

4. Alonso-Lej F, Rever WB Jr, Pessagno DJ. Congenital choledochal cyst, with a report of 2, and analysis of 94 cases. Int Abstr Surg. 1959;108:1-30.
5. Todani T, Watanabe Y, Nurusue M, Tabuchi K, Okajima K. Congenital bile duct cysts: classification, operative procedures, and review of thirty-seven cases including cancer arising from choledochal cyst. Am J Surg. 1977;134:263-9.
6. Lilly JR, Stellin GP, Karrer FM. Forme Fruste choledochal cyst. J Pediatr Surg. 1985;20:449-51.
7. Okada A, Nagaoka M, Kamata S, Oguchi Y, Kawashima Y, Saito R. Common channel syndrome: Anomolous junction of the Pancreato-biliary ductal system. Z Klinderchir. 1981;32:144-51.
8. Sarin YK, Sengar M, Puri AS. Forme Frusta choledochal cyst. Indian Pediatrics. 2005;42:1153-5.
9. Tashiro S, Imaizumi T, Ohkawa H, Okada A, Katoh T, Kawaharada Y, et al. Pancreaticobiliary maljunction: retrospective and nationwide survey in Japan. J Hepatobil Pancreat Surg. 2003;10:345-51.
10. Akhan O, Demirkazik FB, Ozmen MN, Ayuriyek M. Choledochal cysts: ultrasonography findings and correlation with other imaging modalities. Abdom Imaging. 1994;19:243-7.
11. Komi N, Takehara H, Kunitomo K, Miyoshi Y, Yagi T. Does the type of anomalous arrangement of pancreaticobiliary ducts influence the surgery and prognosis of choledochal cyst? J Pediatr Surg. 1992;27:728-31.
12. Sugiyama M, Baba M, Atomi Y, Hanaoka H, Mizutani Y, Hachiya J. Diagnosis of anomalous pancreaticobiliary junction: value of magnetic resonance cholangiopancreaticography. Surgery. 1998;123:391-7.
13. Lenriot JP, Gigot JF, Segol P. Bile duct cysts in adults: a multi-institutional retrospective study-French Association for Surgical Research. Ann Surg 1998;228:159-66.
14. Yamaguchi M. Congenital choledochal cyst: analysis of 1433 patients in the Japanese literature. Am J Surg. 1980;140:653-7.
15. S. R. Banerjee Jesudason, Mark Ranjan Jesudason, Rajiv Paul Mukha, Frederick L. Vyas, Sanjay Govil, John C. Muthusami. Management of adult choledochal cysts: a 15-year experience. HPB (Oxford). 2006;8(4):299-305.
16. Uta Waidner, Doris Henne-Burns, Klaus Buttenschoen. Choledochal cyst as a diagnostic pitfall: a case report. J Med Case Rep. 2008;2:5.
17. Fransisca J. Siahaya, Toar JM. Lalisang, Wifanto S. Jeo, Arnold BH. Simanjuntak, Benny Philippi. Uncommon mixed type I and II choledochal cyst: an Indonesian experience. Case Rep Surg. 2013;2013:821032.

DOI: 10.5455/2349-2902.isj20140813
Cite this article as: Bhasin SK, Azad TP, Digra NC, Mubashar, Sharma G. Missed choledochal cyst: a rare presentation and review of literature. Int Surg J 2014;1:112-5.