

Cystic Dilatation of the Bile Duct (A Case Revealed By a Trauma)

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Abstract: Cystic dilatation of the common bile duct is the most common congenital bile duct abnormality after biliary atresia. It can be discovered at any age, usually accidentally or when it becomes symptomatic but it is exceptionally discovered following a trauma. Only early radical surgical treatment avoids secondary complications, the most serious of which are biliary cirrhosis and cancerization. In our case it was a 4-year-old male who suffered a thoraco-abdominal trauma after being crushed by a cart which surgical exploration had revealed bilious fluid in the peritoneum and perforation of a cystic dilatation of the bile duct type I. After we suture the breach via three stitches and aspiration of the contents of the biliary sac which brought bilious fluid, we proceed at the placement of a drain of Kehr. This was the best thing to do to avoid a serious complication. This is what it's described in literature.

Keywords: cystic dilation, bile duct, post-traumatic.

INTRODUCTION

First described by Vater in 1723, Cystic dilatation of the common bile duct is the most common congenital bile duct abnormality after biliary atresia [1,2]. It can be discovered at any age, usually accidentally or when it becomes symptomatic [5,6] but it is exceptionally discovered following a trauma.

Only early radical surgical treatment avoids secondary complications, the most serious of which are biliary cirrhosis and cancerization [12-15]. The aim of this work is to share the case of a child with cystic dilation of the choledochal post-traumatic revelation which constitutes an exceptional mode of revelation.

MATERIALS & METHODS

We present the case of the 4-year-old male child who suffered a thoraco-abdominal trauma after being crushed by a cart which caused a broken liver, more specifically hepatic segments VII and VIII, extended to the hepatic hile but without lesion of the hepatic pedicle on the abdominal CT scan. The course of action was to monitor him by adopting a non-operative conservative treatment, the child remained hemodynamically stable.

Seven days after his trauma, he presented an abdominal distension with sensitivity to abdominal palpation all moving in a context of fever encrypted at 38.7 °. The control scanner had highlighted a cystic mass appended to the liver with an abdominal effusion

of average abundance, in front of the worsening of the clinical picture of our patient and the CT images we decided to admit to the operating room.

Surgical exploration had revealed bilious fluid in the peritoneum, the gallbladder was of normal size and morphology, and the cystic duct continued with a fusiform cystic mass lined by a peritoneal leaflet (Figure 1) with the presence of a gap in the posterior part of this mass (Figure 2). It was therefore a perforation of a cystic dilatation of the bile duct type I of post-traumatic discovery. Our course of action was to suture the breach via three stitches (Figure 3) after aspiration of the contents of the biliary sac which brought bilious fluid (Figure 4) and the placement of a drain of Kehr (Figure 5). Cystic dilatation of the bile duct was postponed because of the inflammation surrounding the cyst with a high risk of suture release.

The child has kept the drain of Kehr until complete dryness of bilio-cutaneous fistula created by the said drain, currently our patient is doing very well.

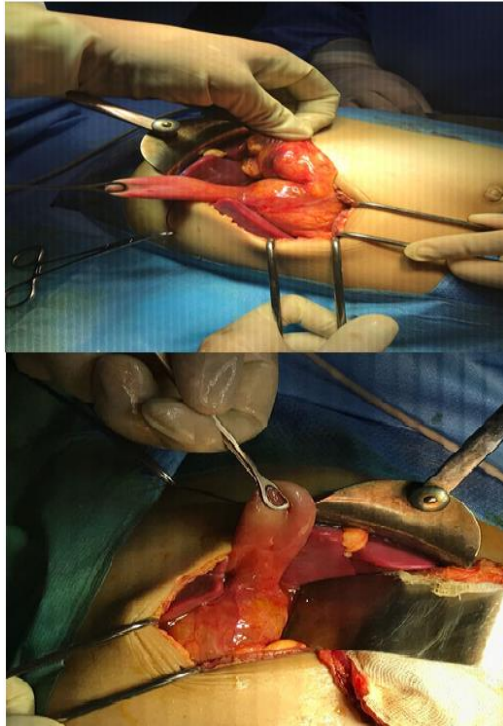


Fig-1: Image revealing bilious fluid in the peritoneum, and the cystic duct continued with a fusiform cystic mass lined by a peritoneal leaflet

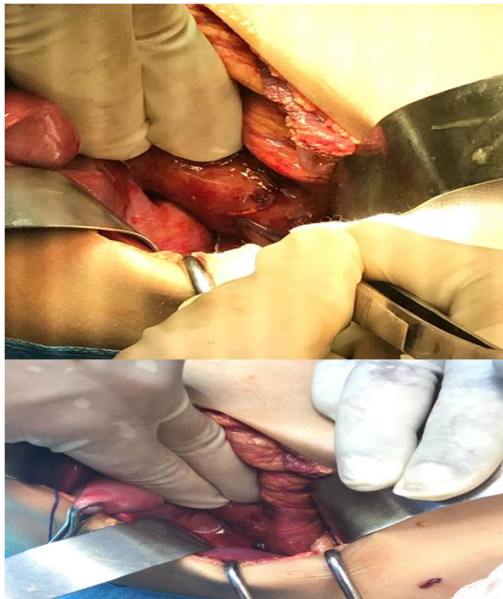


Fig-2: Image revealing the presence of a gap in the posterior part of the cystic mass

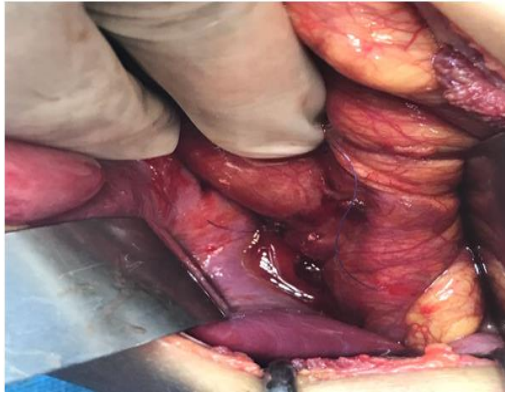


Fig-3: Image showing the suture of the breach

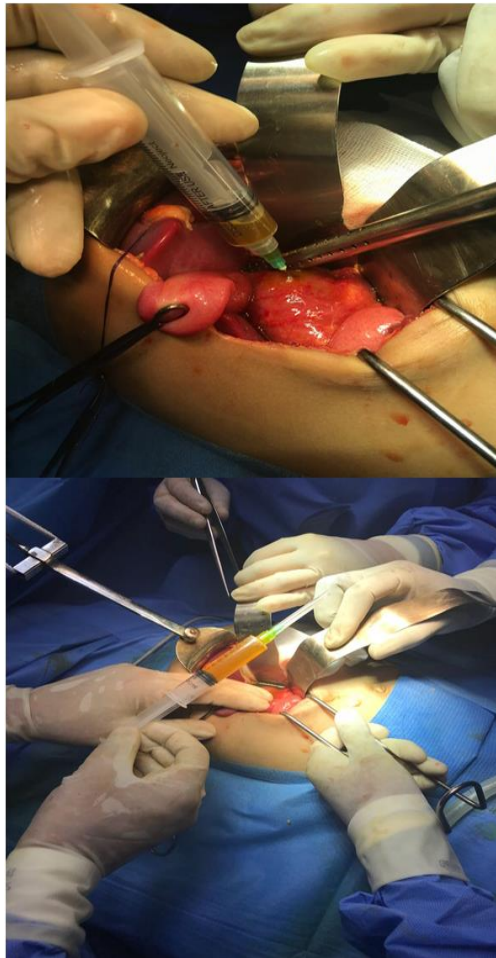


Fig-4: Image showing the aspiration of the contents of the biliary sac which brought bilious fluid

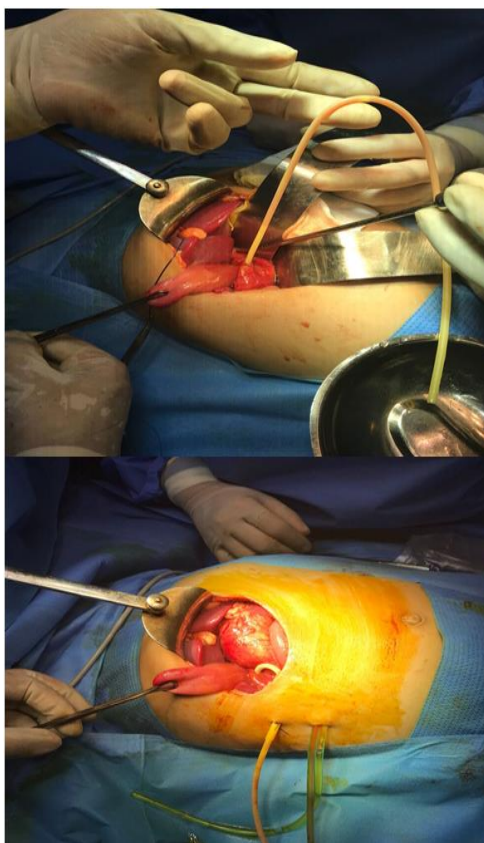


Fig-5: Image showing the placement of a drain of Kehr after the aspiration of the biliary sac

DISCUSSION

Congenital cystic dilatations of the common bile duct are rare malformations: 1/100000 to 1/200000 births per year [7] with a clear predominance in the female sex (70% to 80% of cases) [3,4] which does not corroborate not with our case which is a male subject. They are often found in children, adolescents and young adults, but they occur in 50% of cases before the age of 10 years [5,6].

Bile duct cysts are classified according to their location, extension, and appearance of dilatation. The most common type I involvement that corresponds to the term bile duct cyst is the same type found in our patient. Alonso-Lej *et al.* proposed a classification for extra-hepatic cystic dilatation in 1959, which was modified by Todani incorporating the intrahepatic involvement described by Caroli [10,11].

In terms of etiopathogeny, two theories coexist: that of an embryological defect of the biliary wall, or that of an anomaly of the bilio-pancreatic ductal junction where the Wirsung unites with the bile duct upstream of the sphincter, allowing the reflux of pancreatic juice in the common bile duct [8,9] with a frequency of this anomaly between 10.5 and 58% of cases [7]. The second theory being the most plausible following the work of Babbit.

Clinically, the symptoms begin in 50% to 75% of cases before the age of 10, but these malformations can be discovered in adulthood only and even fortuitously [5,6]. The symptomatic triad, intermittent jaundice, pain, palpable abdominal mass, is very suggestive but only exists in 20% of cases.

Most of the time, only one of the three signs is indicative: jaundice with discrete biological signs of cholestasis is present in 70% of cases, abdominal pain in 60% of cases [16].

Other more exceptional acute complications can be inaugurated namely biliary peritonitis by rupture of the cystic dilatation of the common bile duct (as is the case of our patient), or a haemorrhage by portal hypertension secondary to biliary cirrhosis.

The diagnosis of cystic dilatation of the common bile duct is based on the clinic but also on the paraclinical examinations:

- Ultrasound shows a cystic mass independent of the gallbladder.
- Computed tomography shows a well-defined fluid tumor spread between the portal confluence and the duodenum.

Ultrasonography and computed tomography are also good tests for the diagnosis of intrahepatic abnormalities [17]. The retrograde

cholangiopancreatography is most often necessary before the procedure to detect a complication, to analyze the achievement of convergence and especially to clarify the image of the bilio-pancreatic junction [18]. In our patient we only performed a CT scan in the context of the emergency.

The spontaneous evolution is most often unfavorable: the chronic cholestase and the infection which can lead in a more or less fast time to a secondary biliary cirrhosis. The history can be enamelled of acute complications sometimes mortal, infectious including the repeated or mechanical cholangitis which consists of a traumatic rupture (the case of our young patient) or the spontaneous perforation of the cystic dilation of the bile duct which is the more described in the literature especially in type IV [26].

Cancers of cystic dilation of the common bile duct: Cystic dilation of the common bile duct must be considered precancerous [19, 20, 21]. Surgical treatment is essential and the controversy between supporters of internal drainage and resection is currently closed in favor of the latter. Operative indications vary depending on the type of malformation. For Type I, complete excision of cystic dilatation and gallbladder [22].

In the event of an emergency (acute cholangitis with renal insufficiency or cystic dilatation of the bile duct during an intervention for an abdominal emergency of another nature) this is the case of our patient whom we have taken charge of for abdominal trauma following an accident, immediate excision of the biliary malformation involves a significant risk [23].

In these exceptional cases, it is advisable to perform temporary external biliary drainage that can be performed without major difficulty by cholecystostomy or choledocostomy, then to re-intervene as soon as the patient's condition allows.

However, a study in adults has shown that complicated types of perforation can be operated in one time provided that the patient is stable and in good general condition [24]. A second study on 27 cases of children with spontaneous rupture of cystic dilation of the common bile duct and which highlights the feasibility of performing cyst excision in one time without a two-stage derivation [25], but no publication of the literature deals with the management of post-traumatic perforation of cystic dilatation of the bile duct in a child because the context changes and the trauma in general does not cause the perforation of the cyst in an isolated but induced way other complications of the biliary hepatic junction making the radical surgical cure immediately dangerous. It is for this reason that we opted for drainage in the first place with a surgical cure in two stages.

CONCLUSION

Congenital cystic dilatation of the bile ducts is a rare malformation. His diagnosis is increasingly early thanks to advances in imaging.

Early therapeutic management is therefore necessary to avoid a serious complication, namely spontaneous or post-traumatic perforation that can lead to death.

Conflict of interest

The authors declare that they do not have any conflict of interest and all contributed to the redaction of this publication.

REFERENCES

1. Gillet JY. Échographie des malformations fœtales. 1990. 1re Édition, Paris, Vigot. pp. 173-7.
2. Ka MM, Brissiaud JC, Favre I. Dilatation kystique congénitale du cholédoque: à propos d'un cas diagnostiqué chez une femme sénégalaise. *Dakar Med.* 1992;37:163-6.
3. Cussenot O, Valayer J, Gauthier F. Dilatation congénitale de la voie biliaire principale. *Chir Pediatr.* 1987;28:8-19.
4. Frank JL, Hill MC, Chirathivat S, Sfakianakis GN, Marchildon M. Antenatal observation of a choledochal cyst by sonography. *AJR* 1981;137:166-8.
5. Nagorney DM, McIlrath DC, Adson MA. Choledochal cysts in adults: clinical management. *Surgery* 1984;96:656-63.
6. Todani T, Watanabe Y, Narusue 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.
7. Yamaguchi M. Congenital choledochal cyst. Analysis of 1433 patients in the Japanese literature. *Am J Surg.* 1980;140:653-7.
8. Brunelle F. Pathologie des voies biliaires de l'enfant. *Encyclopédie Médico Chirurgicale.* 1987: A1013.
9. Babbit DP. Congenital choledochal cyst. *Ann Radiol.* 1969;12:231-40.
10. Alonso-Lej F, Rever Jr WB, Pessagno DJ. Congenital choledochal cyst, with a report of 2, and an analysis of 94 cases. *Int Abstr Surg.* 1959;108:1-30.
11. Caroli J, Soupault R, Kossakowski J, Plocker L, Paradowska. Congenital polycystic dilation of the intrahepatic bile ducts; attempt at classification. *Sem Hop.* 1958;34:488-95.
12. Stain SC, Guthrie CR, Yellin AE, Donovan AJ. Choledochal cyst in the adult. *Ann Surg.* 1995;222:128-33.
13. Chijiwa K, Koga A. Surgical management and long-term follow-up of patients with choledochal cysts. *Am J Surg.* 1993;165:238-42.

14. Ishibashi T, Kasahara K, Yasuda Y, Nagai H, Makino S, Kanazawa K. Malignant change in the biliary tract after excision of choledochal cyst. *Br J Surg.* 1997;84:1687-91.
15. Voyles CR, Smadja C, Shands WC, Blumgart LH. Carcinoma in choledochal cysts. Age-related incidence. *Arch Surg.* 1983;118:986-8
16. Florent Ch, Florent M, Flourie B. Les kystes du cholédoque. *Med Chir Dig.* 1986 ; 15 : 405-8.
17. Young WT, Thomas GV, Blethyn AJ, Lawrie BW. Choledochal cyst and congenital anomalies of the pancreatico-biliary junction: the clinical findings, radiology and outcome in nine cases. *Br J Radiol.* 1992 ; 65 : 33-8
18. Baumann R, Uettwiller H, Duclos B, Jouin H, Kerschen A, Adloff M, Weill JP. Dilatation kystique congénitale du cholédoque, anomalie de la jonction biliopancréatique et cancer des voies biliaires. *Gastroentérologie clinique et biologique.* 1987;11(12):849-55.
19. Irwin ST, Morrisson JE. Congenital cysts of common bile duct containing stones and undergoing cancerous changes. *Br J Surg.* 1944; 32: 319-21.
20. Nagorney DM, McIbrath DC, Adson MA. Choledochal cysts in adults: clinical management. *Surgery.* 1984; 96 : 656-63.
21. Bloustein PA. Association of carcinoma with congenital cystic conditions of the liver and bile ducts. *Am J Gastroenterol* 1977 ; 67 : 40-6
22. McWhorter GL. Congenital cystic dilatation of the common bile duct. *Arch Surg* 1924; 8 : 604-26.
23. Moir R, Scudamore H. Emergency management of choledochal cysts in adult patients. *Am J Surg* 1987 ; 153 : 434-8
24. Liwei Chiang, Chan Hon Chui, Yee Low, Anette Sundfor Jacobsen *J Pediatr Surg.* 2018 Apr;53(4):653-655.
25. Ngoc Son T1, Thanh Liem N, Manh Hoan V. One-staged or two-staged surgery for perforated choledochal cyst with bile peritonitis in children? A single center experience with 27 cases *Pediatr Surg Int.* 2014 Mar; 30(3):287-90.
26. Lal R, Agarwal S, Shivhare R, Kumar A, Sikora SS, Kapoor VK, Saxena R. Management of complicated choledochal cysts. *Digestive surgery.* 2007;24(6):456-62.