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Cystic dilatation of the bile ducts: management and long-term results

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Summary
Introduction*: Formerly called as choledochal cyst dilatation in the literature, congenital bile duct dilatation is a rare but not exceptional entity. We aimed to describe the clinical, therapeutic and evolutionary aspects of congenital biliary dilatation in children, based on an experience of 11-years.*
Patients and methods*: Descriptive and retrospective study over 11-years. We collected and analyzed the sociodemographic, clinical and therapeutic data of the patients who were monitored with an average follow-up of 4-years.*
Results*: 15 children were included. The average age was 5-years with a female predominance. Symptomatology was marked by right hypochondrial pain 86.7%, cholestatic jaundice 21%, abdominal mass 28%. This classic triad was complete in 14%. Abdominal ultrasound guided the diagnosis which was confirmed by abdominal CT or bili-MRI. Todani' type I was found in 71% and the type IVa in 28%. The surgical treatment consisted of a total resection of the cyst with a hepatico-jejunal anastomosis on a Roux Y loop for all our patients.*
The immediate postoperative course was simple for 13 patients. We noted one case of anastomotic leakage responsible for a biloma which was drained.The long-term evolution was good with an average follow-up of 4 years.
Conclusion: *Congenital cystic dilatation of the bile ducts is a rare malformation.It can remain asymptomatic for a long time with a risk of degeneration, hence the need for a complete surgical treatment.*
Keywords: bile duct dilatation, congenital, surgery

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I. Introduction:

Formerly referred to in the literature as cystic bile duct dilatation, congenital biliary dilatation (CBD) is a rare but not exceptional entity. 80% of cases are diagnosed in the first decade of life [1]. It is defined as communicating dilatation of the extrahepatic bile duct (EHBD) with or without dilatation of the intrahepatic bile duct (IHBD) [2]. EBVD is classified according to Todani [1] into 5 types. The surgical technique is codified [1,2].

The aim of our study was to describe the clinical, therapeutic and evolutionary aspects of congenital dilatation of the bile ducts in children, based on an 11-year experience.

Patients and methods: This was a descriptive and retrospective study extended over 11 years (January 2011 to January 2021). We included all children aged 0-15 years admitted and managed for CVAD. Other causes of cholestatic jaundice were excluded. We collected and analysed the patients' sociodemographic, clinical and therapeutic data. All patients underwent a primary ultrasound scan supplemented by an abdominal CT and/or biliary MRI. The latter was used to classify the malformation according to Todani [1] and to search for associated biliary-pancreatic junction anomalies. All patients underwent a surgical cure by conventional approach, which consisted of a hepaticojejunal anastomosis (4th jejunal loop). We followed up clinically and radiologically with an average follow-up of 4 years.

II. Results:

During our study period, 15 children were included. The mean age was 5 years [2 months to 15 years] with a female predominance of 12 girls, i.e. a 4:1 ratio. Symptomatology was marked by right hypochondrial pain 86.7%, cholestatic jaundice 21%, abdominal mass 28%. This classic triad was complete in 14%. Other signs were marked by fever, epigastralgia and vomiting. There were two special cases, one with acute

pancreatitis and the other with acute cholecystitis. In the biological work-up, 13 patients had biological cholestasis.

Radiologically, type I represented 10 patients (66.67%) and type IV represented 5 patients (33.33%) (figure 1 and 2). The other types were not encountered in our series. One patient had a pancreas divisum. All patients underwent surgical cure (figure 3 a,b,c). The postoperative course was marked in one patient by an anastomotic leak responsible for a biloma. This was drained with a good evolution.

The average hospital stay was 6 days. The evolution was good in all patients with an average follow-up of 4 years.

III. Discussion:

BVD is a rare anatomical and clinical entity. Its incidence is estimated at 1/13500 births in the USA and 1/1000 births in Asia [3,4]. The epidemiological and clinical data of our patients are in line with those of the literature, particularly with regard to the average age and the clear predominance of women [2,5,6].

As for the radiological type, as in our case, type I is the most common [2]. In the literature, the anomaly of the bilo-pancreatic junction, which has an essential role in the genesis of the anomaly, is reported with a prevalence varying from one author to another, ranging from 30% to 70% [1,2]. No cases were found in our series. However, an endoscopic retrograde cholangio-pancreatography (ERCP) is essential to better ascertain the anomalies of the bilio-pancreatic junction. In the series A. Tannuri et al [6] found that 5 of 7 ERCP patients had a junctional abnormality. S. Mannai et al [2] out of 11 patients, 6 had a junctional defect.

Surgical treatment consisted of complete resection of the HBEV and biliary-gastric anastomosis. For localised Caroli disease, segmental or lobar hepatectomy is performed [2]. Complete resection of the HBEV with biliary-gastric anastomosis was performed in all our patients.

The immediate postoperative complications in our series were a case of anastomotic leakage responsible for a biloma requiring drainage with good evolution. This anatomical fistula is also reported by Salma Foura [7]. and A. Tannuri et al [6].

Other complications reported are lithiasis at the anastomosis, cholangitis, stenosis of the anastomosis, peritonitis, death [2,6,7].

We discuss what to do in the particular case of the fortuitous discovery of a VSD on spontaneous perforation of the cyst resulting in a picture of biliary peritonitis. Is it necessary to operate in one or two stages (peritoneal cleansing with external bypass then a delayed radical cure of the BCD)? Opinions on this case vary according to the series and sometimes within the same series.

T. Ngoc et al [8] in his series of 27 cases, 4 patients were operated on in two stages with a delayed radical cure of 2 to 4 weeks; the 23 patients were operated on in a single stage and he reports a good evolution in all his patients. Our patient was managed in two stages with a delay of 6 weeks. This delay is also variable; A. Ahmed et al [9] reported a 6-month delay between the two procedures. In the long term, the risk of degeneration is not negligible, which requires long-term monitoring [1].

IV. Conclusion:

Congenital cystic biliary dilatation is a rare malformation. It can be asymptomatic or diagnosed in the context of an emergency such as pancreatitis, cholecystitis, biliary peritonitis. The risk of degeneration requires complete surgical treatment with long-term surveillance.

References

[1]. Kevin C Soares, Dean J Arnaoutakis, Ihab Kamel, Neda Rastegar, Robert Anders, Shishir Maithel. Choledochal Cysts: Presentation, Clinical Differentiation, and Management. American College of Surgeons 2014. http://dx.doi.org/10.1016/j.jamcollsurg.2014.04.023.

[2]. S. Mannai, T. Kraïem, L. Gharbi, N. Haoues, H. Mestiri, M.-T. Khalfallah. Les dilatations kystiques congénitales des voies biliaires. Annales de chirurgie 131 (2006) 369–374.

[3]. Howard ER. Choledochal cysts. In : Howard ER, ed. Surgery of liver disease in children. Oxford: Butterworth-Heinemann. 1991.p 78-90.

[4]. Gigot J, Nagorney D, Farnell M. Bile duct cysts : a changing spectrum of disease. J.Hepatobiliary Pancreat Surg 1996;3:405-11.

[5]. Fei Fan, Da-Peng Xu, Zheng-Xiang Xiong, Hai-Jia Li, Hai-Bei Xin, Huan Zhao. Clinical significance of intrapancreatic choledochal cyst excision in surgical management of type I choledochal cyst. Journal of International Medical Research 2018, Vol. 46(3) 1221–1229

[6]. Ana Cristina Aoun Tannuri, Lucas Arjona de Andrade Hara, Guilherme de Freitas Paganoti, Wagner de Castro Andrade, Uenis Tannuri. Choledochal cysts in children: How to Diagnose and Operate on. Clinics. 2020;75:e1539.

[7]. Salma FOURA. Les dilatations kystiques congénitales de la voie biliaire principale chez l'enfant. thèse de doctorat en médecine N° 039 année 2017 Université Cadi Ayad de Marrakech Maroc.

[8]. Tran Ngoc Son, Nguyen Thanh Liem, Vu Manh Hoan. 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) 30:287–290.

[9]. Ahmed Azahouani, Najlae Zaari, Fatine El Aissaoui, Mohamed Hida, Mohamed Fitri, Larbi Benradi. Kyste du cholédoque rompu: revue de la littérature. Pan African Medical Journal. 2019;33:276.

Table I: Comparison of our radiological and therapeutic data with the literature.

Auteurs	**N**	**Type 1 Todani**	**anomalies of the pancreatic trillion junction**	Immediate **Complications**	Long term **Complications**
Our study	15	71%	0	1	0
Tannuri ACA et al.	81	82%	5	3	0
K.C Soares		80-90%	30%		10 à 30%

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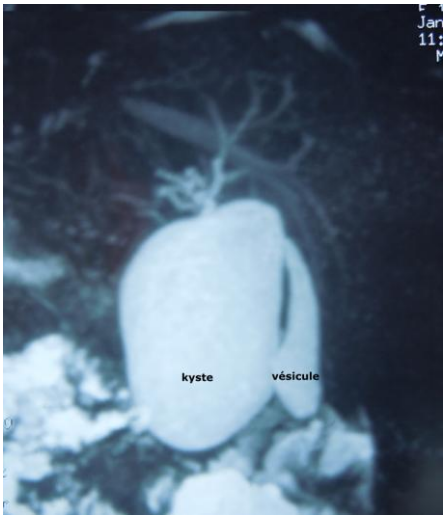

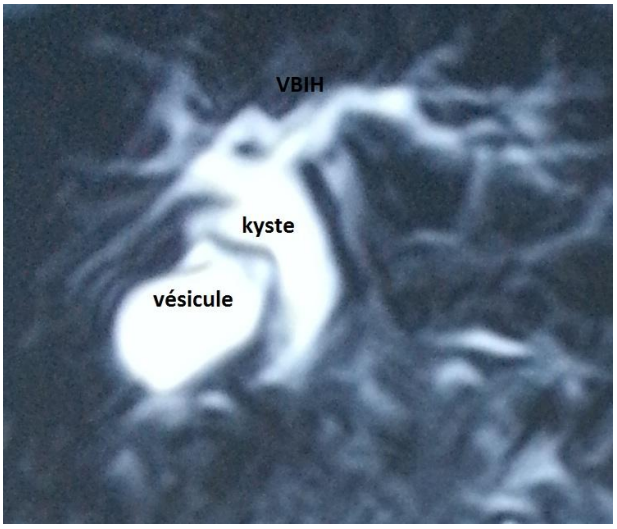

Figure 1 Figure 2

Figure 1 and 2: Bili MRI frontal section images of two of our patients with Todani type I (1) and type IV a (2).

Figure 3: Intraoperative images showing the cyst (a), cyst resection (b) and hepaticojejunal anastomosis (c).

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