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Treatment of choledochal cyst in a pediatric population. A single institution experience of 15-years. Case series

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HIGHLIGHTS

- Choledochal cyst is a rare disease either in pediatrics or adults.
- In the most common type of CC, the surgical excision is the gold standard.
- The reconstruction of the biliary tract in the case of CC is not standardized.
- We present the outcome of our series of patients that either HY or HD were performed.

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ABSTRACT

Background: Choledochal cyst (CC) is a rare congenital anomaly of the bile duct that approximately 75% of the patients are diagnosed in childhood. Without a standardized surgical procedure for the biliary reconstruction, we present our experience over the last 15 years and show the differences between the biliary reconstructions techniques in our population.
Methods: We did a retrospective hospital archive search for patients admitted to the pediatric surgery department with the diagnosis of a choledochal cyst from January 2000 to June 2015.
Results: We found 15 patients, of which, 1 was excluded because of missing data from the hospital record. Of the remaining 14, eight had hepaticojejunal (HY) anastomosis in Roux-en-Y, with a 25% rate of complications; six had hepatoduodenal (HD) anastomosis with a rate of complications of 16.6%. The average hospital length of stay in the group of HD vs. HY was 14 ± 1.6-days vs. 19 ± 8.2-days respectively.
Discussion: There are no standardized surgical reconstruction techniques of the biliary tract after the CC excision, there is literature that supports the biliary reconstruction with an HY and an HD without a distinct advantage over one or the other. Conclusion: In our series HD anastomosis represents a safe procedure with fewer complications than HY.

1. Background

Choledochal cyst (CC) is a rare congenital anomaly of the bile duct (BD) [1], for which there is no standardized surgical treatment [2]. Defined as a pathological dilation of the BD [3,4], if left untreated CC has complications that range from BD obstruction to cholangiocarcinoma [5]. It is estimated that CC has an overall incidence of 1 in 100/150 000 live births in the western population [1], whit a 3–4:1 female predominance, 75% of which are diagnosed in childhood [1,3].

The treatment of the CC will vary accordingly to the Todani's classification (Table 3) [6]; for the commonest type of Choledochal cyst (Type I and IV) the treatment is surgery, although, the reconstruction technique of the BD has no gold standard [7]. The surgical approach has evolved from drainage procedures to the excision of the cyst and reconstruction of the BD as the treatment of choice [5,8]. There are two reconstruction techniques, hepaticojejunal anastomosis in Roux-en-Y and hepaticoduodenal anastomosis, with no apparent benefit of any of them.

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The present report shows the 15-years experience in the management of the choledochal cyst in the Pediatric Surgery Department of the Hospital Civil de Guadalajara "Dr. Juan I. Menchaca". Our primary end-point was to show the differences in the outcome between the reconstructions of the bile duct techniques in our population.

2. Methods

We did a retrospective, hospital archive search for patients admitted to the pediatric surgery department with the diagnosis of a choledochal cyst from January 2000 to June 2015. We did not require the Ethical approval as long as we did not publish any personal data. We found 15 patients, of which, we excluded one because of missing data from the hospital record. A total of fourteen patients were included; we collected the following data: age at the time of presentation, sex, clinical features, associated congenital anomalies, complementary paraclinical tests, surgical technique used for treatment, complications, hospital length-of-stay and following-up.

The statistical analysis was done by parametric statistics with media, average or range for quantitative variables and percentages for qualitative variables. We use Mann–Whitney U-value, Chi-square and Pearson Correlation as needed.

3. Results

We studied a total of fourteen patients (Table 1), 71.42% were female (n = 10), and 28.57% were male (n = 4), the female to male ratio was 2.5:1, the average age of presentation was 5.2-years (range from 1 to 15-years). Clinical symptoms varied in a significant manner, the combination of abdominal pain and jaundice was the predominant symptomatology presented in the 64.2% of our population (n = 9, Table 1). We observed the classic triad of jaundice, abdominal pain and palpable mass only in two patients; also, the association of CC with another congenital anomaly was not seen in our population (Table 1). Two of our patients (patient 8 and patient 9) had a spontaneous perforation of the CC found as an incidental finding during the laparotomy.

Paraclinical tests showed elevated liver enzymes (AST and ALT) in 92.8% of patients (n = 13), as well as elevated alkaline phosphatase in 100%, and 57.1% had hyperbilirubinemia with an obstructive pattern (n = 8). As a diagnostic approach, abdominal ultrasound was performed on all patients, supporting the CC diagnosis in 42.8% (n = 6). Of the regarding eight patients, four had a CT scan, and two had an MRI, achieving the diagnosis of CC in all the six patients. The other two patients were diagnosed during laparotomy secondary to a CC perforation.

The surgical approach consisted of excision of the cyst in all of the patients; the bile duct reconstruction technique was: hepaticojejunal anastomosis in Roux-en-Y (HY) 57.1% (n = 8) and hepaticoduodenal anastomosis (HD) in 42.9% (n = 6). The general complication rate of the two groups was 21.4%; analyzing separately each group, HY had complications in 25% (n = 2/8), and HD in 16.6% (n = 1/6) respectively, this difference was not statistically significant (P = 0.71). The average hospital length-of-stay (LOS) for the HY group was 19 ± 8.2-days vs. 14 ± 1.6-days for the HD group (no statistical significant). The demographic data are described in Table 1.

4. Discussion

Choledochal cyst is a rare entity defined as a cystic dilation of the biliary tract [1–3], its etiology is not well defined, Babbitt DP 1969 [8] theory is the most accepted etiology theory, proposing that an abnormal junction between the biliopancreatic duct (ABPJ) create a shared channel that allows reflux of pancreatic secretion into the biliary tract thus causing increased pressure with subsequent ductal dilation. Further authors have found this association in up to 96% of cases [2,3,9–11]. Other congenital anomalies have been associated with CC like double common bile duct, sclerosing cholangitis, hepatic fibrosis, pancreatic cyst, annular pancreas and cardiac abnormalities [3,12,13]. We did not found any congenital anomaly associated with CC in our series (Table 1); the APBJ was not searched for in our patients.

International literature reports a 3–4:1 female to male ratio with predominance in Asian population [1–3,14], however, in Mexican literature this is variable with different authors reporting different proportions. Orozco-Sanchez et al., 1997 [15] reported a 16:1 ratio, Gallardo-Meza et al., 2010 [16] 2:1, Jimenez-Urueta et al., 2013 [17] 3:1, and Palmer-Becerra et al., 2010 [18] 2.5:1. We found a 2.5:1 female to male relation; nevertheless, after adding all the patients of the Mexican series we found a female to male ratio of 2.8:1 which is close to the international literature. Although the Asian population is more affected than any other ethnicity with an incidence of 1 in 1000 live births [1] Wiseman K et al., 2005 [19] reported the largest North America series of CC and did not found any statistically significant difference in the presentation, management, or outcome between Caucasians and Asians. They also found a female to male ratio of 4:1.

The classical clinical triad of jaundice, pain, and a palpable mass can be found in about 20% of cases, with predominance in childhood [1,3]. We found this triad only in two patients, which makes up for 14.2%. Jaundice and abdominal pain are the most dominant clinical features in children [3,17,20,21], these symptoms were found in our series in 78.5% and 85.7% respectively (Table 2), and together were found in the 64.2% (n = 9, Table 1).

Abdominal ultrasound (US) is the cornerstone of diagnostic approach, whose certainty, can reach a correct diagnosis in 50%, with an appropriate CC classification in 30%, this is directly related to the operator's experience [3,22]. Twelve of our patients had an abdominal US, with an accurate diagnosis and classification of the CC (according to Todani), in 40%. The gold standard to define the anatomy of the biliary tract is the contrast intraoperative cholangiography [23]. Non-invasive test to define the anatomy of the biliary tree include computed tomography (about 90% sensibility and specificity), hepatobiliary scintigraphy with Technetium 99 (HIDA) (67–100% sensibility) and magnetic resonance imaging (MRI) (70–100% sensitivity and 90–100% specificity), they can all be used as diagnostic approach, considering MRI as the first line in preoperative diagnosis to define the biliary tract [1,3,9]. The MRI has the advantage of simultaneously and effortlessly delineates both the biliary and pancreatic duct and therefore accordingly to the cholangiography morphology classify the images ergo Todani's classification [24]. Nonetheless, Acker SN et al., 2013 [25] concluded that preoperative imaging is unable to predict real intrahepatic involvement in CC accurately.

Once the imaging diagnosis is confirmed, we proceed to classify it according to Todanis classification [6] (Table 3); rendering to this classification, a surgical approach is planned, in this case 9/14 our patients had a preoperative diagnosis of CC type I and 3/14 had a preoperative diagnosis of CC type IV, thus treatment consisted of complete excision, cholecystectomy, and biliary tract reconstruction [1,3]; the other two patients had the same procedure once we found the CC intraoperatively. Biliary tract reconstruction technique is not standardized, the systematic review and meta-analysis by Narayanan SK et al., 2013 [26] show that there were no statistically significant difference in biliary leak, cholangitis, anastomotic stenosis, surgical re-intervention and postoperative small bowel obstruction, however there was a difference in biliary reflux favoring HY anastomosis, and a shorter hospital stay in patients who

Table 1
Demographic data of the patients

Patient	Age	Sex	Clinical Symptoms							Todani PreOp	Surgical Technique	Complications	Pathology		LOS days	Following-up
			P	J	V	F	H	PM	T				Biliary Tract	Liver		
1	4 years	F	Yes	No	Yes	No	Yes 2 cm	No	No	1	HD anastomosis	No	Choledochal cyst type 1	NR	14	9 months
2	1 year 8 months	F	No	Yes	Yes	Yes	No	No	No	1	HY anastomosis in Roux-en-Y	No	Choledochal cyst type 1	Microvesicular steatosis	16	6 years
3	1 year 7 months	F	Yes	Yes	No	No	Yes 2 cm	No	No	1	HY anastomosis in Roux-en-Y	No	Choledochal cyst type 1	NR	16	13 years
4	7 years	M	Yes	Yes	No	No	Yes 11 cm	No	No	4	HY anastomosis in Roux-en-Y	POp bleeding	Choledochal cyst type 1	Incomplete septal liver cirrhosis	25	11 years
5	5 years 7 months	F	Yes	Yes	Yes	No	No	No	No	1	HD anastomosis	No	Choledochal cyst type 1	Normal	12	8 months
6	1 year 4 months	F	Yes	Yes	no	Yes	Yes 3 cm	Yes	Yes	4	HD anastomosis	No	Choledochal cyst type 1	NR	14	-
7	2 years 11 months	F	Yes	Yes	Yes	No	Yes 4 cm	No	No	1	HY anastomosis in Roux-en-Y	2011 stenosis of the anastomosis with remodeling of the BR 2013 repetitive grade I cholangitis resolve by antibiotics	Choledochal cyst type 1	Incomplete septal liver cirrhosis	17	5 years
8	3 years	F	Yes	Yes	Yes	Yes	No	No	No	NR	HY anastomosis in Roux-en-Y	No	Choledochal cyst type 1	NR	12	3 years
9	1 year 10 months	M	Yes	Yes	Yes	No	No	No	No	1	HY anastomosis in Roux-en-Y	No	Choledochal cyst type 1	NR	35	3 years
10	15 years	F	Yes	No	Yes	No	No	No	No	NR	HY anastomosis in Roux-en-Y	No	Choledochal cyst type 1	NR	13	2 years
11	2 years 4 months	F	No	Yes	Yes	Yes	No	No	No	1	HD anastomosis	No	Choledochal cyst type 1	Grade 2-3 liver fibrosis	12	8 months
12	15 years	F	Yes	Yes	No	No	No	No	No	1	HY anastomosis in Roux-en-Y	No	Choledochal cyst type 1	NR	15	1 year
13	3 years 8 months	M	Yes	Yes	Yes	No	Yes	Yes	Yes	4	HD anastomosis	No	Choledochal cyst type 1	Moderate chronic cholestatic liver disease	14	1 year
14	8 years	M	Yes	No	Yes	No	No	No	No	1	HD anastomosis	2013 stenosis of the BR 2013 remodeling of the BR with a HY anastomosis in Roux-en-Y	Choledochal cyst type 1	Normal	16	3 years

P=pain, **J**=jaundice, **V**=vomit, **F**=fever, **H**=hepatomegaly, **PM**=palpable mass, **T**=triad, **LOS**=hospital length of stay **PreOp**=preoperative, **NR**=non-referred, **HD**=hepatico-duodenal, **HY**=hepaticojejunal, **CC**=Choledochal cyst, **BR**=biliary reconstruction, **F**=female, **M**=male,

Table 2
Clinical features.

Clinical feature	Patient number	Percentage %
Abdominal pain	12/14	85.7
Jaundice	11/14	78.5
Palpable abdominal mass	2/14	14.2
Vomit	10/14	71.4
Fever	4/14	28.5
Hepatomegaly	6/14	42.8

In our cases series, eight HY in Roux-en-Y were performed, two of these patients presented complications, one had repetitive cholangitis secondary to stenosis of the biliary reconstruction and had to undergo biliary reconstruction remodeling (Table 1), and the other one had a postoperative bleeding that resolve with medical treatment. We did six HD anastomosis with only one patient presenting a stricture of the anastomosis with repetitive episodes of acute cholangitis that required a conversion of the BR into a HY without sta-

Table 3
Modified Todani Classification for choledochal cyst.

I	Cyst limited to the extrahepatic biliary tract. Can be associated with dilation of the intrahepatic duct secondary to biliary stasis.
	IA: Saccular dilation, affecting all or almost all the extrahepatic biliary tree. The gallbladder is originated on the choledochal cyst. Not altered intrahepatic ducts.
	IB: limited saccular dilation, affecting the distal portion of the choledocus.
	IC: fusiform dilation of the common hepatic duct and the choledocus.
II	Extrahepatic supra-duodenal diverticulum, with a normal gallbladder and common hepatic duct.
III	Choledococele: intraduodenal diverticulum
IV	Dilated intra and extra hepatic ducts
	IVA: Fusiform extrahepatic duct dilation and intrahepatic cysts
	IVB Multiple extrahepatic biliary tree dilations, “Pearls Necklace” appearance. No intrahepatic alteration
V	Multiple intrahepatic cysts, no extrahepatic alteration. When combined with congenital hepatic fibrosis, is called “Caroli Syndrome”

underwent HD anastomosis, although there were no randomized clinical trials included in the analysis. We found a shorter hospital LOS in the group of HD, but it was not statistical significant ($P = 0.2$).

tistical significant. In México, three of the five reports on CC support the HD biliary reconstruction technique [16,17,27], Santore MT et al., 2011 [4] and Elhalaby E et al., 2005 [28] sustenance this information in their retrospective comparative series. Nevertheless, it is thought

that there is an increased incidence of biliary reflux with this technique, which also represents an increased risk for cancer development [26,29]. Todani T et al., 2002 [30] reported a case of hilar bile duct carcinoma developing after 19 years following primary cyst excision and HD for biliary reconstruction; similarly, Onu S et al., 2010 [31] reported on their series of 56 patients two patients with biliary tract malignancy after CC resection, one of them developed after 26-year of the initial resection.

The laparoscopic approach for the management of the Choledochal cyst has not a real contraindication, but enough experience of the surgeon with laparoscopic dissection, suture, and vascular control is needed [32]. The intraoperative and postoperative complications are very similar to the open approach and laparoscopic approach, therefore, many centers has applied the laparoscopic management as the routine procedure [33,34].

Our study has several limitations, the study was done in a retrospective matter, we have a small number of patients and a relatively short follow-up of the patients with an average of 3.6-years per patient (ranging from 8-month to 15-year); standardized postoperative follow-up is imperative for long-term conclusions. On the other hand, this represents the experience of a single surgeon in a single institution, which dismissed all possible bias in the surgical judgment. None of the authors had any funding to disclosure.

5. Conclusion

Choledochal cyst is a rare disease with surgical treatment well defined (complete excision). However, the surgical technique for biliary tract reconstruction is not established yet. Until now, evidence shows a small advantage for HY anastomosis [26,35]. Nonetheless, our results although no statistically significant shows an advantage for the HD anastomosis about LOS. Since there are not prospective randomized clinical trials clinical, judgment and experience of the attending surgeon will determine the technique for the biliary tract reconstruction.

Ethical approval

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Author contribution

Hector Silva-Baez: Data Collection, writing.
Pedro Coello-Ramírez: Data Collection, writing.
Eddy Mizraím Ixtabalán-Escalante: Data Collection, writing.
Eduardo Sotelo-Anaya: Data Collection, writing.
Mariana Gallo-Morales: Web search, data collection.
Eduardo Cordero-Estrada: Data Collection, writing.
Victor Hugo Sainz-Escarrega: Data Collection, writing.
César Felipe Ploneda-Valencia: Study design, Data collection, data analysis, writing.

Conflicts of interest

None of the authors declares a conflict of interest.

Guarantor

César Felipe Ploneda Valencia
Hector Silva Baez

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