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Acute Pancreatitis Secondary to a Choledochal Cyst. A Rare Etiology and a Particular Clinical Association

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ABSTRACT

Choledochal cysts (CC) a rare congenital cystic dilation of the extrahepatic and/or intrahepatic bile ducts also named a choledochocele. Approximately 80% of the patients are diagnosed in childhood. Despite the diversity of etiologies, the main elements of the predominantly types I and IV, which constitute the majority of all types, have become clearer. The clinical presentation varies from jaundice in young patients to non-specific abdominal pain in older patients, but morbidity increases with complications such as angiocholitis, pancreatitis, perforation, hepatitis, liver failure and malignancy in late-diagnosed cases. We present a very interested case of choledochal cyst discovered incidental finding following an acute pancreatitis attack.

KEYWORDS

Choledochal cyst; Acute pancreatitis; Common bile duct; Surgical treatment; Hepatico-jejunostomy

1. INTRODUTION

Choledochal cysts are uncommon congenital anomalies of the bile ducts also called a choledochocele. It was first described by Vater and Ezler in 1723, Douglas published the first complete clinical description of the anomaly in a patient in 1853 [1] (Figure 1). He speculated about the congenital nature of this anomaly. In 1959, Alonso-Lej et al. published an extensive review of 94 cases in the literature and added two cases of their own [2]. They classified choledochal cysts into three types. In 1977, Todani et al. further classified this anomaly into five types [3]. Subsequent subtypes based on cholangiographic findings have been described (Figure 2).

The incidence of choledochal cyst is reported to be 1/100000-150000 in western societies, whereas it is 1/1000 in Asian societies [4]. Although choledochal cysts are usually observed in childhood, they are seen in adults in 25 percent of the cases. Many complications can occur before and after surgery in patients with choledochal cyst. It was argued that pre-operational pancreas involvement could be a risk factor for a possible post-operational pancreatitis [5]. The goal of this case is to review the association between choledochal cyst and acute pancreatitis in the literature, and to provide evidence of the possibility of the co-existence of acute pancreatitis with choledochal cyst.

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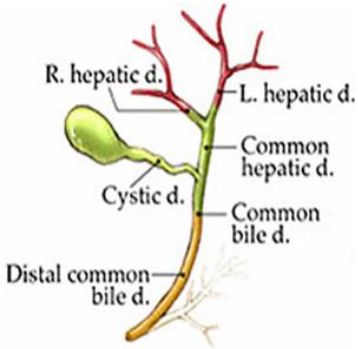

Figure 1: Anatomy of biliary tract.

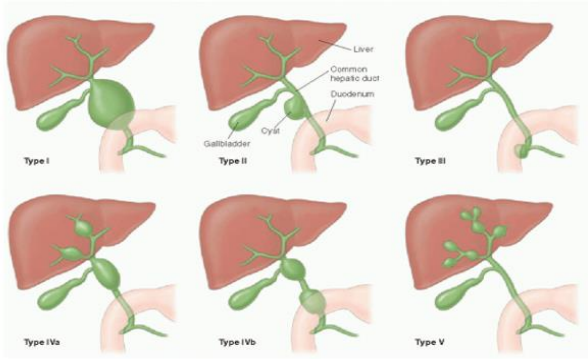

Figure 2: Todani classification of choledochal cysts:
Note: Type I, II and III = Extrahepatic; Type IV = Extrahepatic + Intrahepatic; Type V = Intrahepatic

2. CASE REPORT

A 23-year-old female patient unmarried with no history of any illness admitted for acute pancreatitis. On admission there was no fever T: 37.2°C; heart rate = 60/mn and respiratory rate = 20/mn. Blood investigations reveals WBC = 18000/mm^3. Liver function tests were disturbed with a total bilirubin level of 14 mg/L and an indirect bilirubin level of 10.5 mg/L. The SIRS was <2. The other parameters are within normal limits. Serum lipase = 166.5 IU (more than 4 times the normal value). Abdominal ultrasound examination showed: Voluminous cystic formation of the head of the pancreas (88 mm × 45 mm), no others findings (Figure 3). The CT scan has objectified an aspect of acute pancreatitis with multiple necrosis flows without signs of over-infection classified as stage E by Balthazar. CTSI = 4. Diffuse cystic dilatation of CBD suggestive of a choledochal cyst (Figure 4). The MRCP showed acute pancreatitis with extra-pancreatic necrosis. Choledochal cyst classified TODANI's type I with no sign of degenerescence and a long pancreatico biliary duct measuring 18 mm containing non-obstructive microlithiasis (Figure 5). After a few days of symptomatic treatment the patient underwent surgery. Choledochal cyst excision, cholecystectomy, with Roux-en-Y hepatico-jejunostomy. She passed a smooth postoperative period.

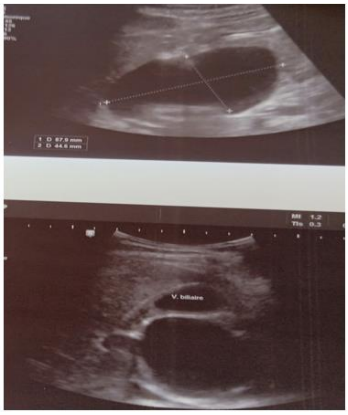

Figure 3: Voluminous cystic formation developing in the head of the pancreas 88 mm × 45 mm the gall bladder is without lithiasis with thin walls.

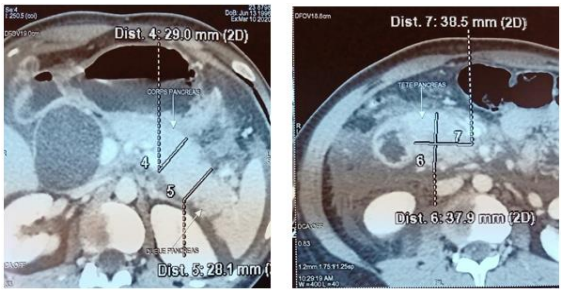

Figure 4: CT showing acute pancreatitis with multiple necrosis flows without signs of over-infection classified as stage E by Balthazar. CTSI = 4, Diffuse cystic dilatation of CBD suggestive of a choledochal cyst.

3. DISCUSSION

Cysts of the bile duct are congenital anomalies of the bile duct and are defined as abnormal and disproportionate cystic dilatation of the bile duct. More than 60% of cases are diagnosed within the first year of life. Prenatal diagnosis has been described, in some cases, as early as 15 weeks' gestation [1]. Choledochal cyst is more prevalent in Asian than in Western countries, and more than 33% of all reported cases are from Japan. There is a female predilection, with a female-to-male ratio of 3:1 -

4:1. The majorities (67% - 80%) of patients have been diagnosed before 10 years of age [6-8]. Symptoms are often non-specific and ambiguous, and clinical outcomes vary with age in cases of bile duct cysts. The three classic symptoms are abdominal pain, a palpable mass in the right upper quadrant, and the presence of jaundice. Adults may present complications more frequently. Jaundice, as well as intermittent biliary obstruction or recurrent episodes of pancreatitis in children, are probable observations [9]. Choledochal cysts are a series of anatomical abnormalities characterized by cystic dilatation in various regions of the biliary tract. The Alonso-Lej classification of choledochal cysts is based on the specific localization of cystic dilatation, refined by Todani et al [3] to include 5 main types of choledochal cysts: type I - type V.

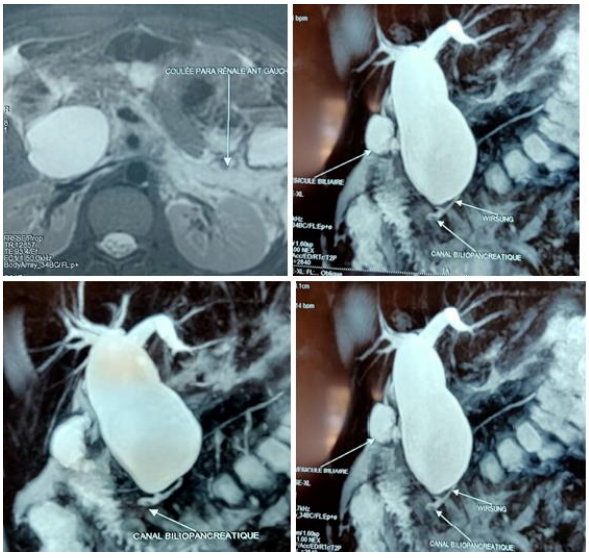

Figure 5: MRCP showed acute pancreatitis with extra-pancreatic necrosis. Choledochal cyst classified Todani's type I with no sign of degenerescence and also a long pancreatico-biliary duct measuring 18 mm containing non-obstructive microlithiasis.

Classification: Alonso-Lej first defined three types of biliary dilatations in 1959; this classification system has been widely accepted ever since. Todani has expanded this classification in 1977 and has subdivided the Choledochal cyst into five subgroups. Todani modified the classification to include anomalies of the pancreatic junction, and the resulting system became the final and most widely used method of classification [10]. According to the Todani classification, Choledochal cysts are classified as follows (Table 1):

Type IA	Cystic dilatation of the extrahepatic bile ducts
Type IB	Extrahepatic distal focal - segmental biliary dilatation
Type IC	Extrahepatic fusiform biliary dilatation
Type II	Extrahepatic biliary diverticula
Type III	Intraduodenal portion of the common bile duct dilatation (Choledococel)
Type IVA	Multiple cystic dilatation of the intrahepatic and extrahepatic bile duct
Type IVB	Multiple cystic dilatation of the only extrahepatic bile duct
Type V	Cystic dilatation of the intrahepatic bile ducts (Caroli's disease)

Table 1: Todani of classification (Type I is the most common with a rate of 75% - 85%).

There are several etiological hypotheses, but the most accepted is Babbitt's theory, which attributes the formation of choledochal cysts to an anomaly in the junction between the pancreatic duct and the main bile duct, outside the Water's ampulla, resulting in a long common duct, which allows the reflux of pancreatic secretion into the main bile duct. Patients with large dilatations of the bile duct have a predisposition to biliary stasis, leading to recurrent cholangitis, calculi development and consequently secondary biliary cirrhosis and malignant transformation [11]. In addition, it is well known that the primary narrowing of the CBD can also contribute to the development of bile choledochal cyst. The types of bile choledochal cysts are determined by the localization, severity and duration of the stricture. Pre-operative recognition of these narrowing is important because the treatment of choledochal cysts without taking the narrowing into consideration can lead to recurrent episodes of cholangitis. This mechanism has been previously underestimated and is now thought to have an even more important role in the physiopathology of the choledochal cyst [12]. In clinical trials, Miyano and Yamataka [13] found an abnormal pancreatico-biliary junction in more than 90% of patients with bile cysts. Abnormal

pancreatico-biliary junction is thought to play an etiologic role in the development of bile duct cysts, particularly those associated with duodenal occlusion [14]. In order to evaluate the pathogenesis of bile-duct cysts, an experimental model of abnormal pancreatic-biliary junction was developed surgically in laboratory dogs; it was shown that varying degrees of dilatation of the common bile ducts developed in 100% of the dogs in 7-10 days, with no further dilatation occurring on subsequent days. Histological analysis revealed signs of chronic pancreatitis in several of these subjects, indicating that an abnormal pancreatico-biliary junction may be an important etiologic factor not only in the formation of bile cysts, but also in the pathogenesis of chronic pancreatitis, a known consequence of bile-cyst disease [15]. The most frequent symptom seen in adults is abdominal pain. In some patients, the symptoms are only very mild. Complications of choledochal cyst are characterized by gallstones (80%), angiocholitis, liver abscess, cirrhosis of the liver, portal hypertension, spontaneous rupture of the CBD and pancreatitis [16-18]. Rarely, patients with choledochal cysts can develop cholangiocarcinoma but the estimated percentage of this malignancy in patients with choledochal cysts has been reported to be as high as 9% - 28%, which is 25-40 times higher than the incidence in the normal population [19,20].

Lal et al. [20] presented a large series of complicated choledochal cysts in 144 patients over 15 years, but with a small incidence of pancreatitis (1.4%). In their analysis, one patient with choledocele had repeated episodes of acute pancreatitis that were successfully managed with trans-duodenal sphincteroplasty, and a second patient was found to have chronic pancreatitis [21]. Another study by Swisher et al. analyzed 32 adult patients who were treated for choledochal cysts. They found 30 documented attacks of acute pancreatitis in 18 patients (56.3%); these occurred in all types of choledochal cysts and were not associated with the age, sex, or race of the patient. They found that the eight patients who had an abnormal pancreatic-biliary junction all developed acute pancreatitis, compared to only two of the six patients with normal pancreatic-biliary duct anatomy ($P<0.006$). In their research, only one patient with a type I choledochal cyst had developed chronic pancreatitis [22]. An abnormal anatomical configuration of the pancreatico-biliary ductal junction (common duct more than 15 mm long) are observed and allowing reciprocal reflux of bile and pancreatic juices into the biliary tree, which can cause inflammation, ectasia and dilatation [23,24]. Amylase and lipase may be found in bile and may explain the high incidence of cholangiocarcinomas and gallbladder carcinomas [25]. In our patient the diagnosis of bile choledochal cyst has never been made before. It was during this first occurrence of acute pancreatitis that the diagnosis of choledocele was known.

In the diagnosis of choledochal cyst, USG is the best method for evaluating intrahepatic and extrahepatic bile ducts, and gallstones. USG is also important in demonstrating complications such as cystolithiasis, cholangitis, and malignancy. In a study, abdominal pain and jaundice were found to be the most common complaints of patients with choledochal cyst. These findings are not particular and therefore the diagnosis is frequently difficult, especially during pregnancy [26]. In our case, we used US & CT first to make a correct diagnosis of acute pancreatitis (Figure 3 & Figure 4). Scanning is not only necessary to demonstrate the continuity of the cyst with the biliary tree, but also to determine the relationship of the cyst with the surrounding structures and the presence of any associated malignancy. In order to adequately plan the operative strategy, CT scan cholangiography can be performed to identify the complete anatomy of the biliary tree, but unfortunately it is less sensitive to visualize the

pancreatic duct which is responsible for the reflux of contrast into the bile ducts [27].

While ERCP has long been considered the gold standard for the diagnosis of bile cysts and the evaluation of abnormalities of the pancreato biliary junction, numerous studies over the past decade have shown that ERCP is as sensitive as or more sensitive than the conventional cholangiogram [28,29]. Since CRMP is a non-invasive study, it avoids some of the significant morbidity associated with ERCP, including the development of moderate to severe acute pancreatitis, which is observed in almost two per cent of patients undergoing ERCP [30]. MRCP has also proven to be as effective as intraoperative cholangiography in planning surgical strategy. Indeed, the lower cost and morbidity compared to other imaging/diagnostic procedures, as well as the reliability of recognition of abnormalities associated with bile-duct cysts, including cholangiocarcinoma and choledocholithiasis, are some of the main properties of PCRM [31]. ERCP should only be performed in situations where an accurate diagnosis cannot be made by other less invasive methods, or where therapeutic competence (complications such as cholangitis or obstruction of gallstones [32,33] and stabilization of patients for the next definitive surgical procedure) is required [34,35].

The diagnosis of acute pancreatitis is based on the sudden onset of typical abdominal pain with at least a three-fold elevation of the upper limit of serum lipase. However, serum lipase may be normal in 20% of cases that are clinically and radiologically confirmed as acute pancreatitis. As observed in our patient diagnosed with a choledochal cyst, elevated serum lipase is not specific and may occur in other diseases of the biliary system, as obstructions of the bile and pancreatic ducts, and choledochal cysts [36]. In our patient the diagnosis of choledochal cyst was discovered incidentally during an investigation of an acute pancreatitis attack with typical epigastric pain and elevated serum lipase (3xnle) and confirmed by CT scan she never presented any symptoms related to her choledochal cyst.

Fusiform-type choledochal cyst is a significant risk factor for preoperative pancreatic complications in choledochal cysts. While postoperative pancreatic complications were relatively rare, preoperative pancreatic complications might be risk factors for postoperative pancreatitis. It should be noted that there is little work in the literature on the pancreatic complications of choledochal cysts, especially in children. In their study, Fujishiro et al. [5] found that preoperative acute pancreatitis was most often observed in children with Todani type I C choledochal cysts. After surgery for choledochal cysts, acute pancreatitis was found in 9.7% of patients with preoperative acute pancreatitis [5]. Despite a plethora of diagnostic procedures, the diagnosis can be difficult to make. Several differential diagnoses have been established. The differential diagnosis of choledochal cyst includes much pathology including biliary atresia, infectious hepatitis, embryonic hepatic rhabdomyosarcoma, biliary lithiasis, acute pancreatitis, biliary hamartoma, and especially biliary atresia, which is one of the two causes of neonatal obstructive jaundice in the neonatal period [37]. It is particularly difficult to distinguish cystic biliary atresia, a subtype of biliary atresia with a totally distinct therapeutic approach, from choledochal cyst. It is therefore essential to establish an accurate diagnosis promptly [38].

The treatment of choledochal cysts is surgical, except for multiple intrahepatic type V cysts, which may benefit from medical management for varying lengths of time [39]. The period of treatment for choledochal cysts diagnosed before birth is still the subject of much debate. Some authors suggest that cysts can be operated on within 2 to 6 weeks, even if asymptomatic, because of the potential risk of complications, while others suggest that they can be followed for a period of time by

ultrasound monitoring and regular assessment of liver function [40,41]. In the past, surgical suction and external drainage were frequently used because the vast majority of patients were pretty sick and a simple and quick procedure was feasible. These methods of external drainage of the biliary tree failed for the most part because of multiple complications, particularly angiocholitis and recurrent biliary fistulas. Mortality was high. The percutaneous drainage surgery can be performed prior to definitive surgery in cases of severe and acute pathology (angiocholitis). This procedure is safe and generally well tolerated; however, it is not necessary in the most patients [42-45]. The treatment of choice is a surgical approach and consists of total removal of the cyst followed by reconstruction with biliodigestive anastomosis. Laparoscopic surgery for choledochal cysts was described in 1995 [46] and has been confirmed to be effective in children as young as 3 months of age and weighing as little as 6 kg [47].

Based on clinical studies, the incidence of acute pancreatitis in children with choledochal cysts was found to be 0% - 70.6%, while in adults it was found to be as high as 10% - 54.5% [48,49]. Protein deposits are also frequent in patients with choledochal cysts. It is well established that reflux of bile into the pancreatic duct affects the distribution of pancreatic secretion and, due to the production of mucin in bile cysts, reflux of pancreatic juice into the bile duct and activation of pancreatic enzymes lead to the formation of protein sediments in bile cysts [50]. Patients with protein formation have been found to be at high risk of pre-operative acute pancreatitis [5]. The choice of treatment varies according to the type of choledochal cyst and the associated hepatobiliary pathology. Type I treatment involves excision of the cyst and a Roux-en-Y hepatico-jejunostomy. For type II, the diverticulum is excised and for type III, excision of the cyst with or without sphincterotomy was performed until very recently. The opening of the bile duct and pancreatic duct in the cyst was an important factor in protecting these ducts in re-anastomosis. Today, endoscopic sphincterotomy and opening of the upper part of the cyst are preferred. In type IV B, as in type I, dilated extrahepatic cysts are completely resected and hepatico-jejunal Roux- en-Y anastomosis is performed. If type V is detected in only one liver lobe, a hepatic lobectomy can be performed. Patients with generalized involvement should be closely monitored for hepatic failure, biliary cirrhosis and portal hypertension. These patients may require liver transplantation [51]. Important advantages of the laparoscopic approach are excellent visualization (view) and less bleeding [52], as well as better postoperative healing, reduced surgical trauma, less postoperative pain, less trauma to the abdominal wall, shorter drainage time from the abdominal cavity, reduced postoperative paralytic ileus and shorter hospital stay. General complications and mortality rates are also lower compared with patients operated on by conventional surgery [53].

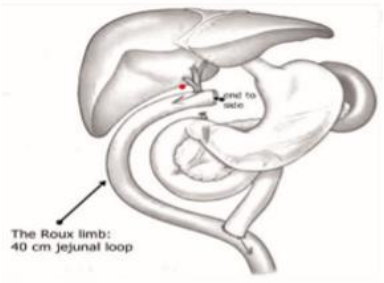

Figure 6: Hepatico-jejunostomy in a Roux-en-Y procedure.

Many other groups have successfully performed assisted laparoscopic ablation and laparoscopic resection of total cysts with Roux-en-Y hepato-enterostomy, with complication rates comparable to those performed in conventional surgery [54]. Li et al. performed laparoscopic resection of cysts with laparoscopically assisted Roux-en-Y hepato-enterostomy in 35 children (33 cyst types I, two fusiform) without conversion to open surgery and with postoperative stays of 3 to 5 days.

The procedure is described in detail by Martinez-Ferro et al (2005) for surgery for bile duct cyst and biliary atresia [55]. Liuming et al. [52]**,** and Liem et al. [53] also concluded that laparoscopic excision was as safe as conventional surgical excision (Figure 6).Our patient was first treated for acute pancreatitis by medical treatment and when the clinical and CT signs (Balthazar's PA stage E) were no longer present, she underwent a choledochal cyst resection with biliary-jejunal anastomosis (hepato-jejunal anastomosis) in Roux en Y. She passed a smooth post-operative period. She was discharged on the seventh post-operative day. She was seen in OPD (Figure 7 & Figure 8).

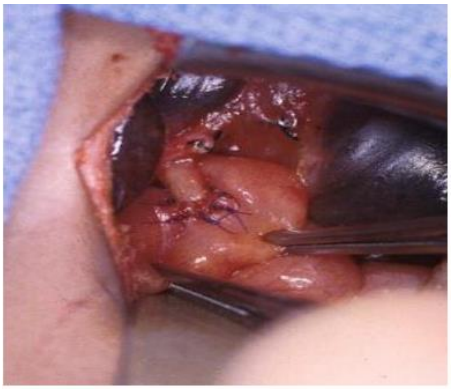

Figure 7: Intraoperative view of a Roux-en-Y hepatico-jejunal anastomosis to re-establish bilioenteric continuity after resection of a type I bile-duct cyst.

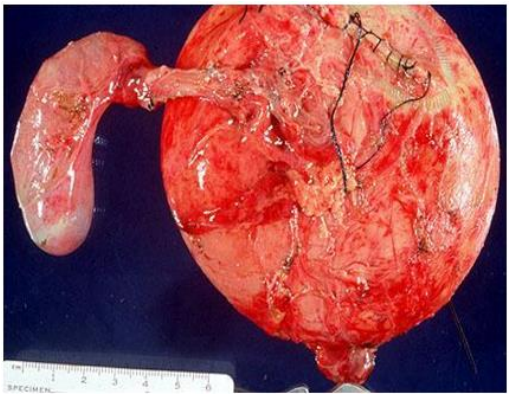

Figure 8: Operative specimen after resection of a type I choledochal cyst.

4. CONCLUSION

Choledochal cyst is a rare condition for which surgery is well accepted (complete excision). It is important to diagnose Choledochal cyst and treat preoperative pancreatic complications by excising the distal canal and completely removing the protein plugs. However, the surgical technique for bile duct reconstruction is not yet well defined. So far, observations show a slight advantage for hepato jejunal Roux Y anastomosis. It should be kept in mind that patients with pancreatic signs, as in our case, are part of the group of patients at risk of developing acute pancreatitis.

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