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Recent Advances in the Diagnosis and Management of Choledochal Cysts

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Abstract

***Aim**: Choledochal cysts are a rare congenital anomaly in which the intra- and/or extrahepatic bile ducts are abnormally dilated. The aim of this study is to provide an updated review on the diagnosis and treatment of these cysts. **Material and method**: A literature search was performed in Medline and Scopus databases to select relevant articles, obtaining a total of 38 articles. An association has been observed with advanced maternal age, overweight of the newborn and birth after the estimated delivery date. **Statistics and Result:** The recommended treatment is resection of the cysts to restore the normal flow of bile into the intestine. Although the prognosis is generally favorable, the risk of malignancy should be kept in mind and periodic check-ups should be performed to detect any signs of malignancy.*

Keywords: *Congenital Anomaly, Dilatation, Bile Ducts, Extrahepatic*

1. Introduction

Common bile duct cysts are a rare abnormality that affects the bile ducts, presenting abnormal dilations in both intrahepatic and extrahepatic bile ducts This condition is characterized by the formation of sacs or pouches that develop along the bile ducts, which can lead to disturbances in the normal flow of bile.

Although considered a rare condition, common bile duct cysts are clinically significant due to the potential complications that can arise, such as biliary obstruction, infection, and gallstone formation (Soares wt al., 2014).

Although Douglas is credited with the initial description of common bile duct cysts in 1852, it is important to note that it was Váter and Ezler who provided a more detailed description of this condition in 1793, laying the groundwork for its recognition and further study (Alonso, 1959). The pioneering work of Váter and Ezler was instrumental in better understanding common bile duct cysts and establishing a solid foundation for future research in this field. Its detailed description allowed a more accurate identification of common bile duct cysts, which in turn has contributed to the advancement of knowledge and understanding of this pathology over the years.

The contribution of Váter and Ezler has allowed to establish a more precise classification of common bile duct cysts and has served as a starting point for further research on this condition. His initial observations have paved the way for the development of more effective diagnostic techniques and therapeutic approaches in the management of common bile duct cysts. It is important to recognize and value the work of these pioneers in the field of common bile duct cysts, since their work has laid the foundations for the current knowledge of this pathology and has been fundamental in improving the diagnosis and treatment of affected patients.

As for the classification of common bile duct cysts, it has evolved over time to achieve a better understanding and management of this condition. The initial classification was proposed by Alonso-Lej in 1959, establishing different types of common bile duct cysts based on their location and pathological characteristics (Alonso, 1959).

However, in 1977, Todani made modifications to this classification, establishing a new classification that has been widely accepted today (Todani et al., 1977). Todani's classification is based on the anatomy and characteristics of cysts, dividing them into five main types (Type I, Type II, Type III, Type IV and Type V), which allows a better understanding of the condition and facilitates the selection of the most appropriate therapeutic approach for each case. Knowledge of the history and classification of common bile duct cysts is critical for healthcare professionals as it provides a solid foundation for the diagnosis and management of this condition. Understanding the historical evolution of common bile duct cysts allows us to appreciate how the knowledge and experience accumulated over the years have contributed to a better understanding and approach to this pathology.

Thanks to advances in the understanding of common bile duct cysts, it has been possible to improve the accuracy in the diagnosis of this condition. Clinical studies and research have made it possible to identify specific characteristics of each type of cyst, which has facilitated the differentiation and precise classification of clinical cases. This is crucial, as an accurate diagnosis is critical to establishing a proper and personalized treatment plan for each patient.

In addition, advances in the understanding of common bile duct cysts have led to the development of more effective and personalized therapeutic approaches. Based on the established classification and understanding of the factors that influence the presentation and progression of cysts, health professionals can design treatment strategies tailored to the needs of each patient. This can range from monitoring and conservative management in asymptomatic cases to more invasive surgical interventions, depending on the severity and specific characteristics of each case.

2. Materials And Methods

To carry out this literature review, we used an exhaustive search strategy in the Medline databases through PubMed and Scopus. We defined the relevant search terms and keywords for each specific section of the study. For this, specific search algorithms were used, such as "choledochal cysts" and "laparoscopy" or "choledochal cysts" and "biliary ducts".

The search was limited to articles written in English, and included only clinical trials, controlled clinical trials, meta-analyses, multicentre studies and reviews. Additional filters, such as "Humans" and "10 years", were applied for sections related to risk factors and pathophysiology of common bile duct cysts.

Articles written in a language other than English and those that did not provide complete information or were not directly related to the subject matter of this review were excluded.

In addition, some articles were included at the discretion of the authors due to their historical relevance in the field of study of common bile duct cysts. After applying the inclusion and exclusion criteria, a total of 38 articles were selected that met the requirements established for this bibliographic review. This rigorous methodology of searching and selecting articles ensures that a thorough and evidence-based review on the topic of common bile duct cysts is obtained, encompassing significant clinical research and relevant studies in the current scientific literature.

3. Results and Discussion

Common bile duct cysts are a relatively uncommon pathology compared to other conditions of the biliary system, but their importance lies in the possible serious complications that can arise if they are not properly diagnosed and treated. These cysts can lead to blockages in the bile ducts, recurrent infections, gallstone formation and even the development of malignant diseases.

It is essential that healthcare professionals and those involved in the diagnosis and treatment of common bile duct cysts are familiar with certain fundamental features of this pathology. This will allow them early detection, an appropriate therapeutic approach and better management of the possible associated risks.

Among the key features to be known is the diversity of clinical and radiological presentations of common bile duct cysts, which makes their diagnosis challenging. In addition, it is essential to have knowledge about the different classifications used to describe common bile duct cysts, such as the Todani classification, which is based on the anatomy and characteristics of the cysts. This helps guide the therapeutic approach and make informed decisions about the management of each individual case.

In addition, it is important to understand the potential complications and risks associated with common bile duct cysts, such as biliary obstruction, infection, and the risk of malignancy. These factors should be taken into account when planning treatment and long-term follow-up of affected patients.

Epidemiology

The incidence of common bile duct cysts is significantly higher in the Asian population, approximately 1/1,000 (Shah et al., 2009) while in the Western population, it is estimated 1/13,000 to 1/150,0005. It is more frequent in the female sex, in a ratio of 3.5:1. 60-80% are detected during childhood and 38% correspond to children under two years of age (Cheng et al., 1996).

Classification

The current classification includes the following subtypes (Figure 1):

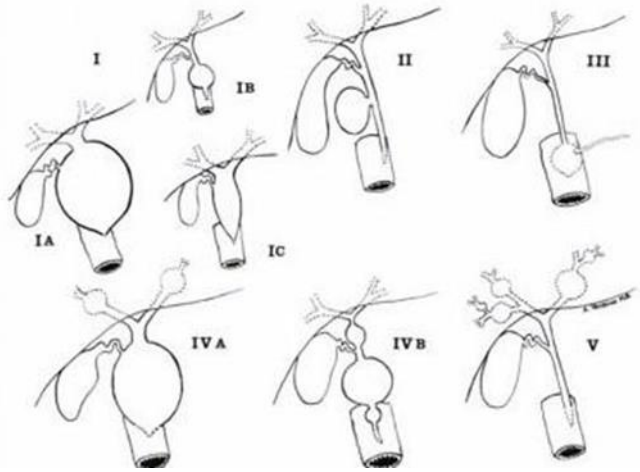

Figure 1. Classification of Todani.

Source: Todani, 1997

Type I. Extrahepatic bile duct dilation: cystic type IA, focal IB and spindle IC

Type II. Diverticulum of the common bile duct.

Type III. Cystic dilation of the intramural portion of the common bile duct or also called coledococele.

Type IV. It is subdivided into two varieties: IVA, involving the intra- and extrahepatic bile duct and type IVB, involving only the extrahepatic route.

Type V. It is located in the intrahepatic bile duct and is characterized by several cystic formations in it, it is also called Caroli syndrome and is generally associated with other malformations such as polycystic kidney (Levy & Rohrmann, 2003).

Sarris and Tsang divide the choledochocele into type A (the common bile duct opens into a dilated cystic segment, which communicates with the duodenal lumen through an independent hole) and B

(the bile duct opens normally to the duodenal lumen and the choledochococele arises as a diverticulum of the common intraampullary canal).

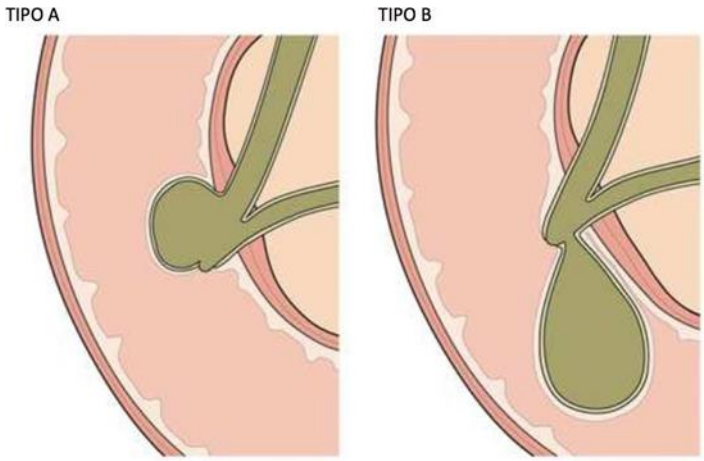

Figure 2. Classification of coledocole

Source: Sarris and Tsang, 2012

Aetiology

The exact etiology of common bile duct cysts is unknown. Several theories have been proposed, among these, the most accepted, is the alteration at the level of the union, between the common bile duct and the pancreatic, distant from the duodenum, which originates a common channel, which would allow the reflux of pancreatic secretions into the bile duct, causing weakness of the wall and formation of cysts. This defect has been found in 57-80% of cases; Therefore, this theory would not explain the origin of cysts in those that do not present this congenital alteration (Makin & Davenport, 2012).

Up to 15% of cases are detected prenatally and during the first two months of life, which would indicate a possible intracystic activation of pancreatic enzymes (Imazu et al., 2001). Another theory proposes, an obstruction of bile outflow. This has been demonstrated in animal models (Park et al., 2014).

Imazu et al. in 2001 proposed as responsible, the dysfunction of the sphincter of Oddi9. Other authors report nodal defects similar to those described in Hirschprung's disease (Kusunoki et al., 1988). Few cases of family association have been reported (Iwata et al., 1988).

Clinical presentation

The clinical manifestations are varied, ranging from no symptoms to developing fatal complications. The classic triad of abdominal pain, jaundice and palpable mass in the right upper quadrant is present in only 20% of cases (Badabarin et al., 2017).

When present in the pediatric population, biliopancreatic junction defect is common, resulting in jaundice and/or abdominal mass; whereas, in adults, this defect is unusual, with abdominal pain being the most frequent symptom (Huang et al., 2010). The risk of developing biliary lithiasis and cholangiocarcinoma is 20% in adults vs. 0.7% in children (He et al., 2014).

Diagnostic methods

The main methods of diagnosis are imaging studies. These include: abdominal ultrasound (AE), computed tomography (CT), endoscopic retrograde cholangiopancreatography (ERCP) and magnetic resonance imaging (MRI), with its variety of study called: hepatobiliary scintigraphy with iminodiacetic acid (HIDA).

Abdominal ultrasound: it is the most used method due to its wide availability. It has a sensitivity of 71% to 97%, and has also demonstrated its usefulness in postoperative assessment (Bhavsar et al., 2012).

Computed tomography: it is useful in presurgical studies, as it allows to delimit the extension of the cysts. In addition, it allows detecting lesions suggestive of malignancy (Lewis et al., 2015).

Hepatobiliary synchography with iminodiacetic acid: allows the detection of spontaneous rupture of cysts (especially in neonates and infants), as well as possible communications with the bile duct (Lee et al., 2009).

Magnetic resonance: it is the method of choice for the detection and assessment of common bile duct cysts, over CT and ERCP. Its sensitivity ranges from 96% to 100%. It has the advantage of clearly delimiting the bile duct, biliopancreatic junction and postsurgical alterations. Unlike ERCP, it poses no risk of perforation, hemorrhage or pancreatitis. The main limitations are: detection of small cysts and minor ductal anomalies (Soreide, 2007).

Endoscopic retrograde cholangiopancreatography: it is reserved for specific cases, such as identification of choledochococces, sphincterotomy and when there are doubts in MRI (oh et al., 2015).

Complications

These include: gallstones and stenosis, cholangitis, cholangiocarcinoma, pancreatitis, secondary biliary cirrhosis and spontaneous cyst rupture (Satkinson et al., 2003).

Up to 10% of the paediatric population has portal hypertension secondary to biliary cirrhosis or portal thrombosis (Chen et al., 2015).

Bile duct neoplasm is the most feared complication; It occurs in up to 30% of adults and is more frequent in the extrahepatic route (50-62% of cases). Other locations are: gallbladder in 38% to 46% and intrahepatic route, in 2.5%. The risk is low in the paediatric population, 0.7% ten years after diagnosis (Tadokoro, 2012).

The risk of neoplasia changes according to Todani's classification: type I has a risk of 70%, type IV 20%, and type III less than 2%. In a study conducted by Sastry et al. evaluated 5,780 patients in the period 1996 to 2010 diagnosed with common bile duct cyst; Of these cases, 434 progressed to neoplasia, equivalent to 7.5%. Of the total cases, only nine equivalent to 0.4% occurred in children under 18 years of age.

The most common cancer was cholangiocarcinoma followed by gallbladder carcinoma. The authors conclude that the risk of malignancy is very low before the age of thirty and from this point it increases with each decade of life24. Cholecystectomy does not reduce the risk of malignancy and once the neoplasm appears, life expectancy is between 6 and 21 months (Ten, 2018).

Currently, from pathology data it is known that neoplasms secondary to common bile duct cysts follow the sequence, hyperplasia - dysplasia – carcinoma (Ulrich et al., 2008).

Treatment

It consists of the resection of the cyst and the proper maintenance of bilioenteric flow.

For the restoration of enteric bilium flow, there are two widely described techniques: hepaticoduodenostomy and Roux-en-Y hepaticojejunostomy. Despite the technical facility and being closer to a physiological condition, a recent meta-analysis showed that hepaticoduodenostomia is associated with greater complications such as reflux and gastritis; Therefore, today, hepatocojejunostomy is the preferred surgical option for most surgeons (Narayanan et al, 2013; del, 2020; Calderon, 2020).

With advances in laparoscopic surgery, open surgery has been displaced. Initially, laparoscopy was used in the pediatric population with excellent results and with very low conversion rates, today its use has also been extended in adults, with similar results.

Comparing laparoscopic and open surgery, the rate of postoperative complications is lower in the laparoscopy group and the rate of bleeding and intraoperative complications are similar in both groups (Yu, 2016; Aly et al., 2018).

The use of monoport has been proposed, demonstrating a success rate similar to the classical technique with four ports, but with better aesthetic results. The drawback is the need to use articulable equipment (Tang et al., 2016).

It is estimated that the learning curve, for laparoscopic resection of common bile duct cysts, are 37 procedures; From here, surgical time, postoperative complications and days of hospital stay decrease (Wen et al., 2017).

The preparation of the bilioenteric anastomosis is the longest step during surgery. The most serious complication, although rare, is leakage of the bilioenteric anastomosis. Its frequency (5%) is similar to that reported in conventional or laparoscopic surgery. This complication can be fatal (Nag et al., 2017).

A recent study compared the use of conventional laparoscopic technique vs. robotic surgery, demonstrating in a statistically significant way the reduction in the number of cases of anastomosis leakage with the use of a robotic platform. The main disadvantages are long time, high costs and additional trained personnel (Lee et al., 2018).

For the treatment of coledococele, the currently preferred technique is endoscopic, since it allows resection and drainage of the same into the intestinal lumen with minimal complications. For type A, endoscopic sphincterotomy is recommended. For type B, partial or total resection of the cyst, after its reduction, by incision or aspiration of the contents. Surgery is indicated in cases where endoscopic therapy fails, the most common procedure being transduodenal excision with or without sphincterotomy (Law & Topazian, 2014).

Liver transplantation is reserved for cases in which the liver is severely affected and with insufficient functional reserve (Cerwenka, 2013).

It is crucial to conduct a thorough investigation of possible extrabiliary congenital affectations in patients diagnosed with common bile duct cysts. A study conducted in the United States revealed a significant association between common bile duct cysts and cardiac anomalies, with up to 31% of cases with this comorbidity detected (Murphy et al., 2012). Despite these associations, overall, the prognosis for patients with common bile duct cysts is excellent, with survival rates reaching 90% at five years.

However, it is important to keep in mind that post-surgical complications can occur after the first thirty days of the intervention. Among the most frequent complications are stenosis of the anastomosis and the formation of intrahepatic gallstones (Takeshita et al., 2014) These complications should be considered and closely monitored during postoperative follow-up to ensure optimal recovery and prevent possible long-term complications.

Importantly, the risk of malignancy in patients with common bile duct cysts persists even fifteen years after surgical excision of the cysts. Studies have revealed that this risk increases progressively over time, being 1.6% at age fifteen, 3.9% at age twenty, and 11.3% at age twenty-five (Ohashi et al., 2013; Blacio, 2020; Lapo, 2020).

These findings underscore the importance of regular monitoring and long-term follow-up in patients who have undergone excision of common bile duct cysts. These controls should include biochemical and imaging tests, such as liver function tests, bilirubin levels, liver imaging studies, and endoscopic retrograde cholangiopancreatography (ERCP). These tests can detect possible recurrences of cysts, changes in liver function and the presence of any indication of malignancy (Estupinan, 2021; Ricardo, 2019; Galves; 2020).

Early detection of malignancy is critical to initiating appropriate and timely treatment, which can significantly improve patient prognosis and survival. In addition, periodic check-ups provide an opportunity to assess the status of liver function and the presence of other complications related to common bile duct cysts.

4. Conclusion

Despite advances in the knowledge of common bile duct cysts, there is still a need to fully understand the genetic mechanisms involved in their development. Although the incidence of this type of cyst is low in western regions, it is crucial to have a thorough knowledge of this disease due to the high mortality rate associated when they are not detected in time and evolve into cancer. The diagnosis of common bile duct cysts is mainly made during childhood, while in adults it occurs incidentally due to variability in its clinical presentation. However, with the current availability of diagnostic equipment and the increase in its use, an increase in the detection of asymptomatic cysts has been observed, allowing for early diagnosis and timely treatment. Importantly, the risk of malignancy persists even after resection of cysts. Therefore, it is crucial to implement periodic surveillance programs to ensure early detection of possible recurrences or signs of malignancy. These follow-up programs, which may include biochemical and imaging tests, are critical to monitoring disease progression and providing timely therapeutic interventions.

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