



## Biliary cysts in adults. 26 years experience at a single center



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### HIGHLIGHTS

- The objective is to analyze prevalence and characteristics of biliary cysts in our center.
- Vague clinical presentation results on a delay of the diagnosis and treatment.
- Surgery is recommended because of increased risk for malignant transformation.
- Most common complication in our patients was stenosis of bilioenteric anastomosis.

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### ABSTRACT

**Background:** Biliary cysts are duct dilatation that can occur on all biliary ducts, 20–25% is diagnosed in adults. The classic triad for the clinical presentation consists on abdominal pain, jaundice and abdominal mass. The standard treatment is surgical resection and bilioenteric anastomosis. The objective of this study is to analyze the prevalence and characteristics of biliary cysts in our center.

**Methods:** This is an observational retrospective study, we included patients older than 16 years old with biliary cyst from march 1989 to February 2015. The demographic and clinical information was collected from the charts and electronic records available at our Hospital.

**Results:** Biliary cysts were reported on 52 patients, only 25 clinical charts were available. The main symptom was abdominal pain in 21 (84%). The diagnosis was performed with abdominal ultrasound in 16 (64%). The most frequent type was IA in 9 (36%). All patients were treated with surgery as a definitive management.

**Discussion:** Vague clinical presentation results on a delay of the diagnosis and treatment. Surgical resection is recommended for patients since they have an increased risk for malignant transformation. Postoperative complications in our patients were stenosis of bilioenteric anastomosis in 3 (12%) patients. **Conclusions:** Biliary cysts require an accurate diagnosis and surgical treatment in order to decrease the risk of malignant transformation and progression of the disease. Precise surgical treatment is needed to achieve complete resection and a long term postoperative follow up is mandatory.

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### 1. Background

Biliary cysts are duct dilatation that can occur on all biliary ducts [1]. These type of lesions are rare and clinical presentation can include a variety of hepatic, biliary and pancreatic diseases [3]. The incidence of biliary cysts is about 1:100,000 to 1:150,000 [4,5], and is more frequent in women [2,6]. Almost 20–25% is not diagnosed until adulthood [6,7] and 51% of the cystic lesions occur on patients older than 20 years old [7,8]. There are several theories that attempt

to explain the etiology of these lesions, the mechanisms that has been described imply obstruction of the distal common biliary duct (CBD) and structural weakness of the duct wall; nowadays the most accepted theory is an abnormal connection between biliary and pancreatic ducts known as abnormal biliopancreatic confluence (ABPC) [3]. On adults we can frequently find inflammatory changes, erosions, few mucin glands and metaplasia [4,12]. Malignant transformation is usually found on the posterior wall [11].

The classic triad for the clinical presentation of biliary cysts consists on abdominal pain, jaundice and an abdominal mass on the upper right quadrant and is present only on 5–10% of pediatric patients and on almost non adult patients [13–15]. Chronic abdominal pain is the main form of presentation (50–96%),

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**Table 1**  
Frequency of biliary cysts, surgical treatment and postoperative complications.

Type	Frequency (%)	Surgical treatment	Complications (%)
IA	9 (36%)	Resection + Cholecystectomy + BDB	3 (33.3%)
IB	4 (16%)	Resection + Cholecystectomy + BDB	2 (50%)
IC	6 (24%)	Resection + Cholecystectomy + BDB	0
II	1 (4%)	Resection + Cholecystectomy + BDB	1 (100%)
III	1 (4%)	Cystectomy + BDB	1 (100%)
IVA	4 (16%)	BDB (Choledocho-duodenum) Resection + Cholecystectomy + BDB Liver surgery + Resection + Cholecystectomy + BDB	2 (50%)
IVB	0	NA	NA
V	0	NA	NA
TOTAL	25 (100%)	—	9 (36%)

BDB: biliodigestive bypass surgery.

followed by jaundice and cholangitis. Other symptoms include nausea, vomit, weight loss and gastrointestinal bleeding. The diagnosis of biliary cysts is made frequently based on findings on ultrasound or CT scan, which have a sensibility of 70–97% [16]. The MR with colangiopancreatography is the best non invasive method for diagnosis of biliary cysts [5,10]. When cysts or dilatation are identified, a CT or an ERCP should be performed to determine the extent of the disease and the presence of malignant lesions, since this disease is associated with cholangiocarcinoma with an incidence of 10–30% [17]. Patients with previous biliary surgery have the same risk of malignant transformation as patients without this type of procedure [18]. Because of this risk of malignant transformation for biliary cysts type I, II and IV, the standard of treatment is surgical resection with cholecystectomy, resection and hepaticojejunostomy [3], a partial hepatectomy may be needed if there are intrahepatic cysts that are consider resectable [19,20].

The objective of this study is to perform a retrospective analysis of patients in our center with diagnosis of biliary cysts in order to know the prevalence and characteristics of this disease in our population.

## 2. Methods

This is an observational retrospective study, we included all patients older than 16 years old with confirmed diagnosis of biliary cyst referred and treated at our institution, Centro Medico Nacional Siglo XXI, from march 1989 to February 2015. The demographic and clinical information was collected from the charts and electronic records available at our Hospital.

## 3. Statistical analysis

The analysis was performed with SPSS v 23 software for the descriptive statistics, frequencies and central tendency measures, we also performed a chi square test to calculate the distribution and differences between the available variables.

## 4. Results

Biliary cysts were reported on 52 patients, of which only 25 clinical charts for information were available. From this population 20 (80%) were females and 5 (20%) were males, with a mean age of 31.3 years old. The main symptom was abdominal pain in 21 (84%), followed by jaundice in 13 (52%), nausea and vomiting 9 (36%), pancreatitis in 4 (16%), cholangitis in 2 (8%), weight loss in 2 (8%), and abdominal mass in 1 patient (4%). Only one patient had the classic presentation of jaundice, abdominal pain and abdominal mass.

The diagnosis of biliary cysts was performed with abdominal ultrasound in 16 (64%), ERCP in 11 (44%), MRI in 10 (40%), CT scan in 9 (36%), cholangiography in 1 (4%) and Scintigraphy in 2 patients (8%).

The most frequent type of biliary cysts in our population is IA in 9 (36%) patients, IB in 4 (16%), IC in 6 (24%), II in 1 (4%), III in 1 (4%), and IVA in 4 (16%).

All patients were treated with surgery as a definitive management. Patients with biliary cysts type IA, IB, IC and II had a resection, cholecystectomy and reconstruction with Roux-en-Y anastomosis (Table 1). Patient with type III had a previous surgery with a Roux-en-Y anastomosis and had a biliodigestive bypass surgery. Type IVA cyst patients had liver surgery and a biliodigestive bypass surgery.

**Table 2**  
Complications in patients with biliary cysts treated with surgery.

Type	Surgery	Type of complication
IVA	Resection + Cholecystectomy + BDB	Abdominal sepsis and reintervention.
IB	Resection + Cholecystectomy + BDB	Bleeding (hemoperitoneum) and reintervention.
IVA	BDB (Choledocho-duodenostomy)	Stenosis and reintervention. Carcinoma.
II	Resection + Cholecystectomy + BDB	Stenosis and reintervention.
III	Cystectomy + BDB	Bleeding (hematoma), stenosis, leakage; reintervention.
IB	Resection + Cholecystectomy + BDB	Bleeding (hemoperitoneum) and reintervention. Anastomosis dehiscence and reintervention. Biliary fistula
IA	Resection + Cholecystectomy + BDB	Biliary fistula (conservative treatment).
IA	Resection + Cholecystectomy + BDB	Biliary and pancreatic fistula.
IA	Resection + Cholecystectomy + BDB	Pulmonary embolism. Leakage and reintervention.

BDB: Biliodigestive bypass surgery.

Postoperative complications in this population were abdominal sepsis in 1 (4%) patient and bleeding in 3 (12%), whom required reintervention. In 3 (12%) patients we founded stenosis of the bilidigestive anastomosis and 1 (4%) developed a biliary fistula, both required later reintervention. One patient with a type IVA cyst transformed in malignant disease and was treated with surgical resection (Table 2). All patients were discharged and follow up was performed as outpatient.

## 5. Discussion

In our study, abdominal pain resulted on the most frequent form of presentation of biliary cysts (84%), as describe on previous studies like Edil et al. [1]. On the other hand, Wiseman [9] reported an unspecific presentation in adults and this often results on a delay of diagnosis and treatment, since the classic triad of pain, jaundice and abdominal mass occurs only in 1–52% of the patients. In our group of patients, weight loss, cholangitis and pancreatitis were less common and the classic triad was only identified in 1 (4%) patient. This vague clinical presentation results on a delay of the diagnosis and treatment of this group of patients because a high level of suspect is needed, in our center, the average time between the reference to our hospital and surgical treatment is 21.7 days (SD  $\pm$  45 days).

Surgical resection is recommended for patients with biliary cysts since they have an increased risk for malignant transformation. In our study most of patients with biliary cysts was treated with resection with bilioenteric anastomosis and 1 (4%) of them had a report of carcinoma, which shows the presence of malignant transformation of this pathology and the risk for young patients to have a carcinoma, also this patient had postoperative complications such as stenosis attributed to the oncologic diagnosis. Postoperative complications in our patients were stenosis of bilioenteric anastomosis in 3 (12%) patients, but survival is 100% in our group, which shows the importance of early diagnosis and accurate surgical treatment that results in a good prognosis.

## 6. Conclusions

Biliary cysts require an accurate diagnosis and surgical treatment in order to decrease the risk of malignant transformation and progression of the disease. Most of the patients with dilatation or cysts can be treated with surgical resection and bilioenteric reconstruction. Precise surgical treatment is needed for biliary cysts to achieve complete resection and a long term postoperative follow up is mandatory for this patients because of the risk of developing malignant transformation. In our center the standard of treatment is surgical resection with bilioenteric anastomosis and long term follow up with adequate outcomes for our patients.

## Ethical approval

Approved by the local research committee on 22/06/2009, reference R-2009-3601-98.

It is an observational study, no interventions were done to the patients in this study.

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## Author contribution

All authors contributed equally to this paper.

## Conflicts of interest

The authors declare no conflict of interests.

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## Guarantor

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## Consent

Written informed consent was obtained from the patient for publication of this report.

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