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Biliary cysts in adults: Cerrahpaşa experience

Ali Vedat Durgun[1], Sefa Ergün[1], Başar Can Turgut[1], Osman Şimşek[1], Mehmet Velidedeoglu[1], Kaya Sarıbeyoğlu[2], Salih Pekmezci[1]

[1] Department of General Surgery, İstanbul University-Cerrahpaşa Cerrahpaşa Faculty of Medicine, İstanbul, Türkiye
[2] Department of General Surgery, Charite University, Berlin, Germany

ABSTRACT

Objective: Biliary cysts are biliary duct dilatations, with 20% of the cysts being diagnosed in adulthood. Abdominal pain, jaundice and palpable abdominal mass are defined as the classical triad. However, nausea, vomiting, fever, itching and weight loss are frequent complaints. There are several treatment options depending on the type of the cyst. This study aimed to share our experience with biliary cysts and contribute to the literature on this subject.

Material and Methods: Thirty patients, who received treatment for biliary cyst from January 1981 to December 2018 at our clinic, were studied retrospectively. The patients were analyzed based on age, sex, type of the cyst, diagnosis and treatment methods, post-op follow up and complications.

Results: Twenty-seven of the patients were females, and three were males. The patients were aged between 16 and 76 years, and the median age was 41.9 years. All patients presented with abdominal pain, which was accompanied by cholangitis in nine patients, nausea and vomiting in four patients, dyspepsia in three patients and palpable mass in one patient. According to the Todani classification, biliary cyst findings were consistent with Type I in 23 patients, Type V in three patients, Type IV in two patients, Type II in one patient and Type III in one patient.

Conclusion: Diagnosis and treatment are complex in biliary cysts due to anatomical proximity and variations. Therefore, it would be beneficial to refer them to referral centers. Choice of treatment should be based on the type of the cyst.

Keywords: Biliary cysts, choledochal cysts, Todani, surgical treatment

INTRODUCTION

Biliary tract cysts are surgical issues typically encountered in newborns and children; however, approximately 20% of the cases are diagnosed as late as adulthood. These lesions are usually referred to as choledochal cysts; however, biliary tract cyst is a more suitable term as the cystic dilatation can develop at any part of the biliary tree and it is not specific to the main bile duct (1,2).

Classification system for extrahepatic biliary tract cysts was first proposed by Alonso-Lej, F. in 1959. This classification was revised by Todani in 1977 to include intrahepatic cysts, and the same author updated the system in 2003 based on an anomalous pancreaticobiliary junction (APBJ) case (1,2).

Todani identified five essential biliary cysts, and cystic dilatation of the cystic duct was added in recent years as Type VI classification (3,4).

The incidence rate of biliary cysts is reported to be between 1/100.000 and 1/150.000. The rates are higher at the south and east of the continental Asia (1/1000), and 1/2-2/3 of the reported cases are from Japan. There are around 4000 patients reported up to date. Biliary cysts are more common in females (80%), and they can be seen in women three to eight times more compared to men. Past cases were seen predominantly in children; however, in new cases, the rates in children and adults are remarkably close (1,5,6).

There are different theories on the formation of biliary cysts. The most widely accepted theory is anomalous pancreaticobiliary junction (APBJ). Anomalous pancreaticobiliary junction (Babbitt theory) is seen in 50-80% of the patients with biliary cysts. This is usually seen together with a long common duct facilitating reflux of the pancreatic juice into the biliary tract (5,7). As the ductal junction is outside the duodenum wall and therefore it is devoid of the sphincter of Oddi, it causes reflux of the pancreatic juice into the biliary tree (7).

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Corresponding Author
Sefa Ergün
E-mail: sefaergn@yahoo.com

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Most biliary cyst cases show symptoms and are identified before the age of 10. In 20% of the cases, the complaints do not emerge until adulthood (>16 years). Classical presentation includes the triad of abdominal pain, jaundice and palpable abdominal mass, and it is more frequent in children compared to adults, and most of the patients present only with one or two findings of the triad. Recurrent pain in the epigastric or right hypochondrium region, abdominal tenderness and mild jaundice are the most common onset symptoms (1,5,8).

Cysts are usually suspected during transabdominal US (ultrasound) or CT (computed tomography) imaging of a patient with abdominal pain, jaundice or an abdominal mass. Imaging methods to be utilized for the assessment of biliary cysts are transabdominal US, CT, MRCP (magnetic resonance cholangiopancreatography), ERCP (endoscopic retrograde cholangiopancreatography), EUS (endoscopic ultrasound) and hepatobiliary scintigraphy (HIDA). MRCP has been more commonly preferred in recent years as it is non-invasive (5,8).

The purpose of our study was to draw attention to biliary cysts due to their rare frequency and contribute to the literature on this issue.

MATERIAL and METHODS

Thirty patients who underwent surgery due to biliary tract cysts between January 1981 and December 2018 were included in the study. The data was retrospectively studied using medical records of the patients. The patients were analyzed based on age, sex, type of the cyst, diagnosis and treatment methods, post-op follow up and complications, and the topic was studied in line with the relevant literature.

All quantitative data were expressed as mean ± standard deviation. Qualitative variables were defined by frequencies (%). This study was approved by the Ethics Committee of İstanbul Universty Cerrahpaşa, Cerrahpaşa Medical Faculty in 17.06.2020 with number 83045809-60401.02.

RESULTS

Twenty-seven of the patients were females, and three were males. The patients were aged between 16 and 76 years, and median age was 41.9 years. Twenty-one patients were diagnosed with biliary cysts before surgery. The remaining nine patients were hospitalized with different diagnoses including jaundice, choledocholithiasis, hydatid cyst and were diagnosed during the surgery. All patients presented with abdominal pain accompanied by cholangitis in nine patients, nausea and vomiting in four patients, dyspepsia (fever, pain, jaundice) in three patients and painful palpable mass in one patient.

According to the Todani classification, choledochal cysts findings were consistent with Type I in 23 patients, Type V in three patients, Type IV in two patients, Type II in one patient and Type III in one patient. Roux-en-Y hepaticojejunostomy reconstruction was performed in 16 patients who underwent cholecystectomy + cystectomy due to Type I choledochal cysts, and continuity was ensured in five patients with hepaticoduodenostomy. One patient with Caroli syndrome limited to the left lobe of the liver received left hepatectomy + cholecystectomy + choledochal T-tube drainage. Of the two patients with Type V solitary liver cyst, one received partial cystectomy + cystoraphy + T-tube drainage, and the other patient with localized cyst in segment III ductus and choledoch stones received segment III resection + choledochotomy + T-tube drainage. One of the patients who previously underwent drainage received cystojejunostomy, and four patients received cystoduodenostomy procedures.

US and CT were more commonly used for diagnosis in early patients (11 patients) and recent patients received US followed by a MRCP (20 patients). Pre-op ERCPs were performed for 12 patients for diagnosis and treatment purposes; however, one of the procedures failed. No other preoperative concomitant anatomical anomalies were noted. Two patients who developed post-operative intra-abdominal abscess and collection underwent percutaneous drainage, and two patients with wound site infection were treated with wound dressing.

One patient, who had undergone cholecystectomy + choledocoduodenostomy and developed recurrent cholangitis due to anastomosis stricture in the long-term follow-up, and underwent Roux-en-Y hepaticojejunostomy. One patient who underwent cholecystectomy + cystectomy + Roux-en-Y hepaticojejunostomy in 2004 developed afferent loop syndrome in 2006 and underwent segmental jejunal resection + reanastomosis procedures. One patient who had been operated with cholecystectomy + cystectomy + Roux-en-Y hepaticojejunostomy developed incisional hernia after one year.

Mean hospitalization period was 10.5 days, and mean follow-up period was 8.1 years. No malignancies were observed in the pathological specimens.

Choledochal cyst was discovered in one patient who was taken into surgery for cholelithiasis at another center; the surgery was terminated without any intervention. The second operation was performed in our clinic, and the patient's Type I choledochal cyst was resected successfully.

In another case, a 40-year-old male patient had biliary cyst accompanying gallbladder cancer. Gallbladder cancer (T1-stage according to the TNM clasiffication) was incidentally identified in the cholecystectomy piece; gallbladder bed excision and lymphadenectomy were planned for the patient. A type 1 biliary cyst was diagnosed during the preoperative imaging, and it was resected concurrently. No other malignancies were observed in the pathological examination.

One patient, who was diagnosed with Caroli disease based on the imaging studies due to abdominal pain and cholangitis, had stone of the left intrahepatic biliary tracts and mechanical jaundice. Percutaneous transhepatic catheter (PTC) and biliary drainage was performed pre-operatively, and then the patient received left hepatectomy + cholecystectomy + T-tube drainage. The patient later received percutaneous drainage for post-operative intra-abdominal abscess and was discharged on day 17.

After the introduction of hepatopancreatobiliary surgery in 2000, our clinic abandoned drainage procedures, and performed the resection procedures for all cases, and usually preferred Roux-en-Y hepaticojejunostomy for the reconstruction. Since this study included a large group of patients going back to 1981, some surgical operations that we do not prefer today were also included in the series.

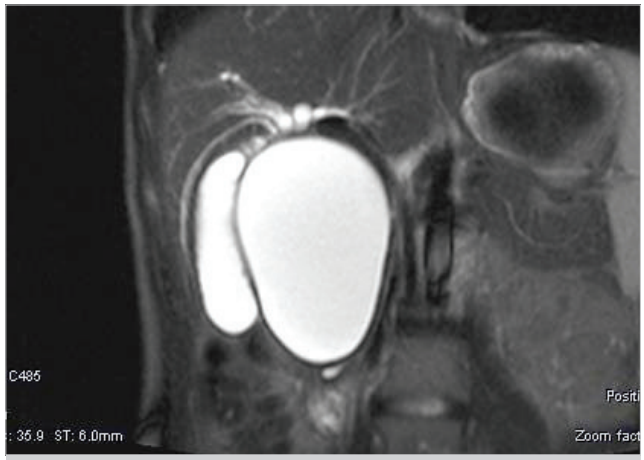

Figure 1. MRCP demonstrating extrahepatic biliary duct dilatation in a Type I cyst.

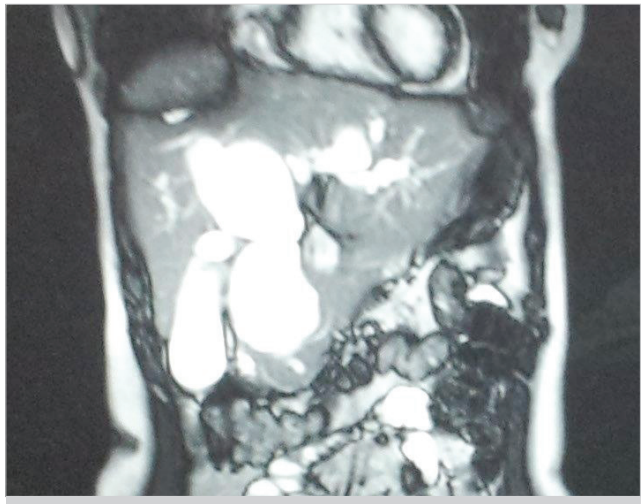

Figure 2. MRCP appearance of extrahepatic and intrahepatic biliary duct dilatation in a Type IVA cyst.

DISCUSSION

The classical presentation of biliary cysts includes the triad of abdominal pain, jaundice and palpable abdominal mass, and it is more frequent in children compared to adults. Most patients usually present with one or two findings of this triad; but nausea, vomiting, fever, itching and weight loss are also among the common complaints (9,10).

The patients may present to the hospital with signs and symptoms of biliary cyst complications such as pancreatitis, cholangitis and obstructive jaundice. Although rare, another presentation is acute rupture of the cyst with subsequent bile peritonitis (5). Nine of our patients presented with cholangitis during diagnosis, seven of which were treated with medical therapy, and the remaining two were operated after biliary drainage with ERCP and PTC for concomitant mechanical icterus.

Advances in the imaging methods have contributed greatly to biliary cyst diagnosis and classification. Cystic lesions are mostly identified by US or CT first. Ulltrasound sensitivity differs between 70% and 97%. The best non-invasive imaging method for biliary tract cysts is MRCP. ERCP and PTC give us detailed anatomical information. ERCP can effectively show the relationship between the pancreatic duct and biliary duct. PTC can be preferred when ERCP is inconclusive in intrahepatic and proximal biliary ducts. MRCP was performed for all 20 patients, the records of whom we could access (Figure 1, 2).

Biliary cysts should be differentiated from pancreatic, mesenteric and hepatic cysts (simple cyst, hydatid cyst) which are not connected to the biliary tract. If there is still suspicion after CT and MRCP, hepatobiliary scintigraphy (HIDA) or ERCP can be performed to establish the connection with the biliary tract (5,8).

Biliary cysts can be mistaken for hydatid cyst in endemic regions. This was the issue of our previous study (11). We focused on this issue after we found a solitary intrahepatic cyst extending outside the surface of the liver in a patient who was taken into operation with hydatid liver cyst diagnosis.

The treatment approach in biliary cysts depends on the type of the cyst. Type I, II or IV cysts are generally eligible for surgical resection. Type I and IV cysts are treated with total excision and Roux-en-Y hepaticojejunostomy or hepaticoduodenostomy (1,8).

Type II cysts are eligible for simple excision. Cholecystectomy may also be used. T-tube is placed if there is a defect on the common bile duct after the excision. Type III cysts are treated with sphincterotomy or endoscopic resection if they are symptomatic. Type V cysts are harder to treat; they require prevention against recurring cholangitis and sepsis. The cysts which are limited to one lobe are treated with right or left hepatectomy, and patients with widespread cysts are transplantation candidates (5,8).

Cases with ascending cholangitis should be treated with antibiotics and bile drainage regardless of the type of the cysts (ERCP or PTC). Of the 21 patients who underwent cystectomy for type I or IV choledochal cysts; 16 received Roux-en-Y hepaticojejunostomy, and hepaticoduodenostomy was preferred for five patients (Figure 3).

Type V cysts may be solitary or multiple. They are referred to as Caroli cysts in most resources (1). This is true for multiple intrahepatic cysts; however, existence of solitary intrahepatic cysts should also be taken into consideration. There were two cases that met this definition in our case series. The treatment method for solitary cysts can be determined based on localization. Of the two cases that met this definition, one received left lateral segmentectomy due to the cystic dilatation of segment III duct and the stones in it (Figure 4). The other case with exophytic growth from the liver surface received partial cystectomy and choledochal T-tube drainage.

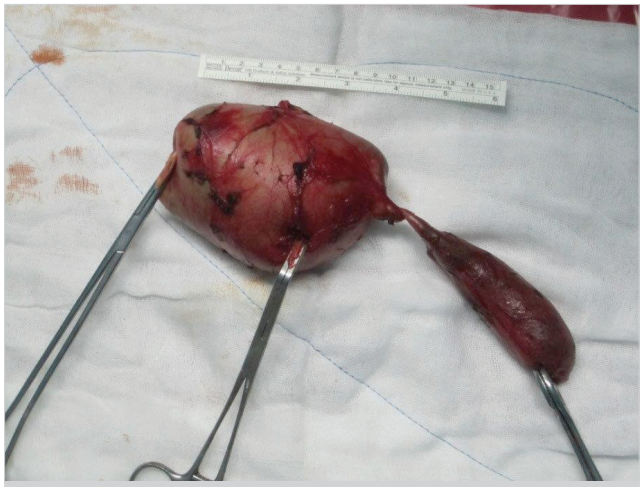

Figure 3. Resection of the Type I biliary cyst.

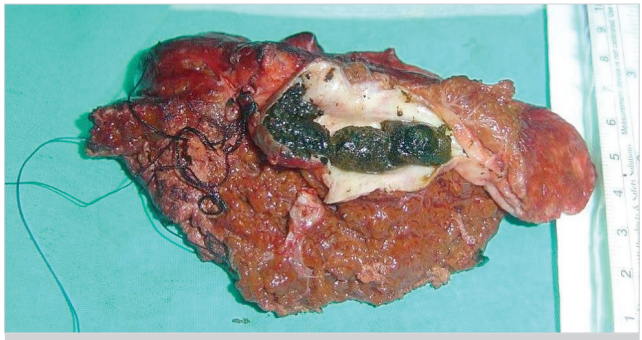

Figure 4. Segment 3 resection of the Type V solitary cyst.

In the current literature, it was recomended that in multiple intrahepatic cysts (Caroli), lobectomy should be performed if the disease is limited to one lobe, and if it is widespread, the recommendation is to treat the cholangitis episodes (antibiotics, PTC) first, and then perform liver transplantation (1,5). There was one patient with multiple intrahepatic biliary cysts (Caroli) in our case series. The patient received left hepatectomy as the disease was localized in the left lobe (Figure 5).

In biliary cysts, the whole bile tree is under the risk of cholangiocarcinoma due to APBJ and pancreaticobiliary reflux. Removal of the cyst does not eliminate future risk of cholangiocarcinoma development (7,12,13).

We did not encounter any cholangiocarcinoma cases in our series.

As for the classical knowledge that biliary cysts carry a risk of cholangiocarcinoma development, we would like to underline that biliary cysts are most common in the Far East and Southeast Asia. Sastry et al. have demonstrated that cancer developed in 7.5% of patients with biliary cysts (70.4% cholangiocarcinoma, 23.5% gallbladder cancer and 6.1% other cancers) (12,14).

Tyson and El-Serag have reported that the rate of malignancy transformation in biliary cysts is higher in Asians (18%) versus Americans (6%). It is also known that biliary tract parasites (*Clonorchis sinensis*, *Opisthorchis viverrini*) are commonly seen in those areas (12,13). In these cases, the risk of cholangiocarcinoma development is certainly associated with concurrent parasites (chronic irritation) rather than biliary cysts.

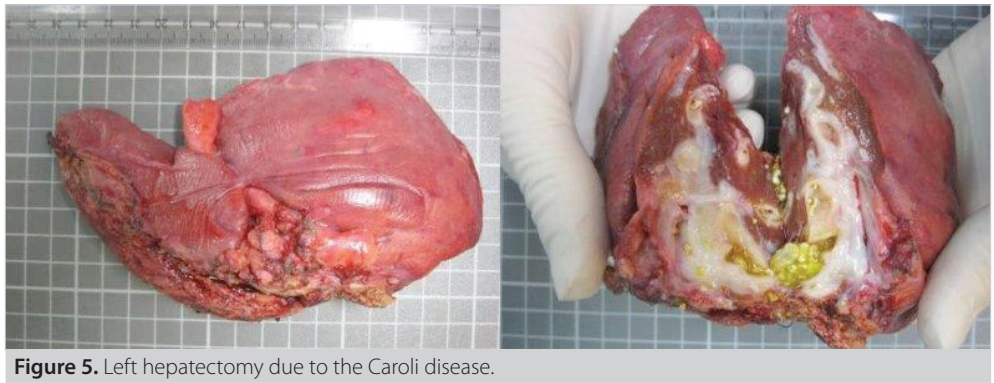

Figure 5. Left hepatectomy due to the Caroli disease.

By their nature, biliary cysts may include some anomalies that could be considered as a complexity for surgeons. Concurrent ductal and vascular anomalies increase the difficulty of surgery. They can be more troublesome when they are detected during surgery rather than preoperative tests (15,16).

An interesting revelation of our series was that some of the cases were not diagnosed with choledochal cysts before surgery. If surgeons with limited experience in this area unexpectedly detect choledochal cysts during surgery, they may choose outdated options for treatment of choledochal cysts. Procedures such as choledocoduodenostomy, cystojejunostomy and T-tube drainage that we came across in our series should be considered within that context.

Hepaticoduodenostomy has been associated with higher gastric and biliary cancer due to bile reflux. Furthermore, meta-analyses comparing RHYJ and hepaticoduodenostomy have found more reflux and gastritis in hepaticoduodenostomy. A wide anastomosis facilitating bile flow to the intestine and reducing anastomotic stricture and bile reflux after cyst excision may prevent complications and development of carcinoma in intrahepatic ducts (8,17).

As long as there are no contraindications, choledochal cysts are resected in order to prevent development of malignancies and complications in the future.

CONCLUSION

In conclusion, biliary cyst diagnosis and treatment should be referred to specialized, advanced facilities as complications are frequent in this area due to anatomical proximity and variations. In Type I cysts, which is the most common subgroup, surgical methods of choice are total cyst resection and RY hepaticojejunostomy.

Ethics Committee Approval: This study was approved by Cerrahpaşa Faculty of Medicine Deanery Clinical Researches Ethics Committee (Decision no: 83045809-60401.02., Date: 07.07.2020).

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Conflict of Interest: The authors have no conflicts of interest to declare.

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ORİJİNAL ÇALIŞMA-ÖZET
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Erişkinde biliyer kistler: Cerrahpaşa deneyimi

Ali Vedat Durgun[1], Sefa Ergün[1], Başar Can Turgut[1], Osman Şimşek[1], Mehmet Velidedeoglu[1], Kaya Sarıbeyoğlu[2], Salih Pekmezci[1]

[1] İstanbul Üniversitesi-Cerrahpaşa Cerrahpaşa Tıp Fakültesi, Genel Cerrahi Anabilim Dalı, İstanbul, Türkiye
[2] Charite Üniversitesi, Genel Cerrahi Anabilim Dalı, Berlin, Almanya

ÖZET

Giriş ve Amaç: Biliyer kistler, safra kanallarında oluşan genişlemeler olup %20 kadarı erişkin yaşta tanı almaktadır. Biliyer kist olgularında karın ağrısı, sarılık ve palpabl abdominal kütle, klasik triyad olarak tanımlansa da hastaların büyük kısmında, bu triyadın sadece bir ya da iki öğesi saptanır. Bulantı, kusma, ateş, kaşıntı ve kilo kaybı sık görülen şikayetlerdir. Tedavide kistin tipine bağlı olarak farklı seçenekler bulunmaktadır. Çalışmamızda biliyer kist olgularındaki deneyimimizi okuyucuyla paylaşmak ve bu konudaki literatüre katkı sunmak amaçlanmıştır.

Gereç ve Yöntem: Kliniğimizde Ocak 1981 ve Aralık 2018 yılları arasında biliyer kist nedeniyle tedavi uygulanan 30 hasta retrospektif olarak incelendi. Hastalar, yaş, cinsiyet, kist tipi, tanı ve tedavi yöntemlerine, ameliyat sonrası takip ve komplikasyonlarına göre analiz edildi.

Bulgular: Hastaların 27'si kadın, üçü erkekti. Yaş aralığı 16-76 olup median yaş 41,9 idi. Yirmi bir hastaya biliyer kist tanısı ameliyat öncesinde konulmuş olup, diğer dokuz hasta mekanik ikter, koledokolithiyazis, hidatik kist vb. tanılarla yatırılmış ve ameliyat sırasında tanı almışlardır. Hastaların hepsinde karın ağrısı şikayeti mevcut idi, dokuz hastada kolanjit bulguları, dört hastada bulantı, kusma, üç hastada dispepsi, (ateş, ağrı, sarılık) bir hastada ağrılı palpabl kütle eşlik ediyordu. Todani sınıflamasına göre 23 hasta Tip I, üç hasta Tip V, iki hasta Tip IV, bir hasta Tip II, bir hasta Tip III biliyer kist ile uyumlu idi.

Sonuç: Biliyer kistler, tanısı ve tedavisi komplike olgulardır. Anatomik komşuluklar ve varyasyonlar nedeniyle ameliyatlarda zorluk derecesi yüksektir. Bu nedenle referans merkezlerine yönlendirilmelerinde yarar vardır. Tedavi şeklinin seçiminde kistin tipine göre davranmak esastır.

Anahtar Kelimeler: Biliyer kistler, koledokal kistler, Todani, cerrahi tedavi

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