Original Article

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Our Surgical Experience in Anomalous Opening of the Common Bile Duct §

Koledok Açılım Anomalilerindeki Cerrahi Deneyimlerimiz

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ABSTRACT

Objective: *The treatment of anomalous opening of the common bile duct opening is performed by endoscopic or surgical methods. In addition to endoscopic procedures, indications of surgical treatment, surgical procedures and complications related to these methods are aimed to be presented in the light of the literature.*
Method: *Patients who underwent surgical treatment for anomalous opening of the common bile duct between November 2014 and August 2018 were analyzed retrospectively. Demographic characteristics, presenting symptoms, diagnostic methods, treatment procedures, complications and postoperative follow-up information were analyzed.*
Results: *A total of 8 patients were included in the study. The mean age was 64.3 years. Male, and female patients constituted 75% (n=6) and 25% (n=2) of the study population. The most common presenting symptoms were abdominal pain (50%) and biliary colic (25%). Cholestatic enzymes were found to be high in 75% of the patients. Fifty-one patients had hyperbilirubinemia. Ectopic biliary drainage was most commonly (75%) located at the bulbus and the second most common (25%) localization was the post-bulbar region. The most common surgical procedure was hepaticojejunostomy (62.5%) and choledochoduodenostomy was performed in 25% of the cases.*
Conclusion: *Surgical treatment methods can be used in addition to endoscopic methods in the treatment of anomalous common bile duct opening. Caution should be exercised in patients undergoing surgical treatment for anastomotic leakage, duodenal ulcer bleeding and bleeding from the area of sphincterotomy.*

Keywords: *choledochus, ectopic opening, surgical treatmant*

ÖZ

Amaç: *Koledok açılım anomalisinin tedavisi endoskopik ya da cerrahi yöntemler ile yapılmaktadır. Endoskopik girişimlerin yanısıra cerrahi tedavi endikasyonları, uygulanan cerrahi yöntemler ve bu yöntemlere ait komplikasyonların literatür eşliğinde sunulması amaçlanmıştır.*
Yöntem: *Kliniğimizde Kasım 2014 ile Ağustos 2018 tarihleri arasında koledok açılım anomalisi nedeni ile cerrahi tedavi uygulanan olgular retrospektif olarak analiz edilmiştir. Hastaların demografik özellikleri, başvuru semptomları, tanı yöntemleri, tedavi prosedürleri, komplikasyonlar ve postoperatif dönemdeki takip bilgileri analiz edildi.*
Bulgular: *Çalışmamıza toplam 8 hasta dahil edildi. Ortalama yaş 64,3 idi. Erkek cinsiyet %75 (n=6), kadın cinsiyet %25 (n=2) idi. En sık başvuru semptomu karın ağrısı (%50) ve bilyer kolik (%25) idi. Kolestaz enzimleri hastaların %75'inde yüksek bulundu. Yüzde elli olguda ise hiperbilluribinemi mevcuttu. Ektopik bilyer drenaj en sık (%75) bulbus, ikinci sıklıkta ise (%25) postbulber bölgede idi. En sık uygulanan cerrahi işlem (%62,5) hepatikojejunostomi, %25 olguda ise koledokoduodenostomi idi.*
Sonuç: *Koledok açılım anomalisi tedavisinde endoskopik yöntemlerin yanısıra cerrahi tedavi yöntemleri de uygulanabilmektedir. Cerrahi tedavi uygulanan hastalarda anastomoz kaçağı, duodenal ülser kanaması ve sfinkterotomi alanından kanama açısından dikkatli olunmalıdır.*

Anahtar kelimeler: *koledok, ektopik açılım, cerrahi tedavi*

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INTRODUCTION

The common bile duct enters the second part of the duodenum from the side of the medial wall and opens into the ampulla of Vater. It may open to the third or fourth part of the duodenum and, less frequently, to the bulbus, stomach or pylorus [1-4]. Anatomic anomalies related to the opening of the common bile duct into the gastrointestinal tract have been further diagnosed through the widespread use of Endoscopic Retrograde Cholangiopancreatography (ERCP). The etiology of anomalous opening of the common bile duct is unknown and considered to be the result of an error during embryonic development of the biliary system [5].

The ectopic opening of the common bile duct to the duodenum was first described by Vesalius in 1543. In this case, one of the double bile ducts opened to the gastric antrum and the other to the duodenum [6]. In the largest case series in the literature, ectopic biliary drainage anomaly was reported in 53 of 12,158 cases who underwent ERCP (0.43%) [1].

Although congenital anomalies of the common bile duct are more common in childhood, they can also be diagnosed in adulthood. While anomalous opening of the common bile duct in adulthood can be identified incidentally, they can also be diagnosed based on symptoms such as biliary stone, recurrent attacks of cholangitis, duodenal ulcer and/or duodenal obstruction [7].

It was thought that the risk of bleeding and perforation was high in endoscopic procedures due to the defective sphincter structure and lack of intramural part of CBD leaning to the duodenum. Therefore, surgical treatment was adopted as the primary treatment modality. Nowadays, with the development of endoscopic intervention technologies and increasing experience on the part of endoscopists, endoscopic treatment methods have gained more importance [1,8,9].

In this article, we aimed to present our experience with patients who underwent surgical treatment due to anomalous opening of the common bile duct in the light of the literature.

MATERIAL and METHOD

Eight patients who underwent surgical treatment at the General Surgery Clinic of Çukurova University Faculty of Medicine due to anomalous opening of the common bile duct between November 2014 and August 2018 were included in the study. Ethical approval numbered 88/16 and dated 05.03.2019 was received from the Çukurova University Faculty of Medicine Ethics Committee. Patient files and hospital information system records were examined and a database was created. The cases were analyzed retrospectively. Follow-up data was supported with telephone interviews with the patients. Patients under 18 years of age and patients whose records could not be reached were excluded from the study.

Patients' demographic characteristics, Body Mass Indexes (BMIs), American Society of Anesthesiologists (ASA) scores, presenting symptoms, preoperative amylase and bilirubin levels, previous abdominal surgery, and diagnostic methods, treatment procedures, intraoperative, and postoperative complications, postoperative hospital stay, re-operations, postoperative 90-day mortality, 90-day unplanned re-admission and mean follow-up times were analyzed.

Endoscopic Retrograde Cholangiopancreatography (ERCP) was performed using the standard technique with the aid of therapeutic duodenoscope (Olympus, TJF 240, Japan) with the patient in the prone position. Anomalous opening of the common bile duct was defined as the absence of a papilla or papilla-like structure in normal anatomical localization in the second part of the duodenum and inability to obtain cholangiogram when the contrast agent was given from the orifice in the ampulla [6]. Sphincterotomy was not performed in cases with choledocholithiasis accompanied by ectopic biliary drainage due to a high risk of perforation.

Endoscopic examination revealed cases with gastric and/or duodenal ulcer, duodenal deformity or stenosis, and these cases were recorded.

Anastomotic leakage was defined as the deterioration of anastomotic integrity detected by clinical and radiological imaging methods.

Wound infection was defined as superficial or deep surgical site infection on the incision site according to the definition of Centers for Disease Control and Prevention (CDC) [10].

Unscheduled re-operation was accepted as a surgical intervention under general, spinal or epidural anesthesia within 30 days after primary surgery, according to the definition of the National College of Surgeons National Surgical Quality Improvement Program (ACS-NSQIP) [11].

Hydration and IV antibiotherapy were applied to patients with cholangitis at the first admission. After determining the etiology of cholangitis, the treatment strategy was redefined.

All patients were treated with low- molecular weight heparin, prophylactic antibiotherapy and compression stockings during induction of anesthesia as antithrombotic therapy.

Follow-up

In the early follow-up after the patients were discharged, they were followed up at in the outpatient clinics for certain periods in terms of wound location and other postoperative complications. In addition to laboratory tests, imaging methods, if available, were preferred according to the symptoms. Subsequent follow-ups were carried out at 6-month intervals in the first year and at yearly intervals after the 2nd year.

Surgical Technique

The patients were operated under endotracheal general anesthesia, with the patient in a slightly inverse Trendelenburg position. After insertion of a central catheter, nasogastric tube, urinary catheter, and skin preparation, right subcostal incision was made. Hepatoduodenal ligament was dissected in the Roux-N-Y hepaticojejunostomy technique. After visualization of the common bile duct from the hilar region to the periampullary region, the right and left hepatic ducts were cut from their bifurcation and distal parts. The distal common bile duct stump was sutured and closed. The jejunum was cut with the help of a stapler, 40 cm distally from the Treitz ligament. Retrocolic anastomosis of the distal jejunum at the distal 40th cm was made with the bifurcation of the posterior wall with continuous 5-0 prolene sutures, while the anastomosis of the anterior wall was made with 5-0 prolene sutures with individual hepaticojejunostomies. Jejenojejunal anastomosis was performed manually, double-layered side by side with 3/0 vicryl and 3/0 silk sutures at the distal 60th cm l. In the portoenterostomy technique, common bile duct was excised above the bifurcation and small bowel anastomosis was performed similarly. Anastomosis of the liver capsule was performed with 6-0 prolene sutures to form a Roux-N-Y portoenterostomy. In the technique of choledochoduodenostomy, the duodenum was mobilized by the Kocher maneuver and the common bile duct was released. Common bile duct was opened with vertical incision then duodenum was opened transverse incision. The lumen of the common bile duct was irrigated. Stone was extracted in patients with choledocholithiasis. Then, anastomosis of choledochoduodenostomy was completed with 4-0 prolene sutures. Gastroenterostomy was performed by the Bilroth II procedure with 3-0 vicryl and 3-0 silk sutures placed retrocolically on the posterior aspect of the stomach. Silicone drains were placed in the Winslow and Douglas cavities in all patients.

Statistical Evaluation

Data were analyzed using IBM SPSS Statistics for Windows, version 24 (IBM Corp., Armonk, N.Y., USA). Categorical variables were summarized as numbers and continuous variables as mean and standard deviation (median and minimum-maximum, where necessary).

RESULTS

A total of 8 patients were included in the study. The mean age was 64.3 years. Male , and female patients constituted 75% (n=6) and 25% (n=2) of the study population. The mean body mass index was 27.6 and the mean ASA score was 2. The most common comorbid diseases were diabetes mellitus (75%) and hypertension (75%).

The most common symptoms were abdominal pain (50%) and biliary colic (25%), but there were also patients presenting with jaundice and fever. Cholestatic liver enzymes were found to be high in 75% of the patients. Leukocytosis was detected in

62.5% of the patients. Fifty percent had hyperbilirubinemia. Four patients had a history of cholecystectomy and one patient had a history of liver resection due to a benign disease.

As concomitant diseases at diagnosis, CBD stone was detected in 5, GB stone in 2, duodenal ulcer in 5 and gastric ulcer in 1 patient. Ectopic biliary drainage was most commonly located at the bulbus (75%), and the second most common localization (25%) was the post-bulbar region (Table 1).

Table 1. Demographics and clinics of patients.

	Mean	Median	Range	Minumum-maximum
Age (years)	64.3	64,5	44	43-87
BMI	27.6	27.65	17,93	22.67-40.6
Sex				**n**
Male				6
Female				2
ASA				
1				0
2				6
3				2
4				0
Comorbidities				
Diabetes Mellitus				3
Hypertension				3
Asthma				1
Coronary artery disease				1
Clinicalcharacteristics Symptoms				
Abdominal pain				4
Biliary pain				2
Icterus				1
Fever/chills				1
Laboratory findings				
Leukocytosis				5
Jaundice				4
Increased serum ALP-GGT				6
Previousabdominalsurgery				
Cholecystectomy				4
Liver resection				1
Concomitantdiseases at diagnosis				
CBD stone				5
GB stone				2
Duodenal ulcer				5
Gastric ulcer				1
Ectopic biliary drainage to				
Bulbus				6
Post bulbar				2

ALP: Alkaline phosphatase, GGT: Gamma-glutamyltransferase
CBD: Common bile duct, GB: gallbladder

Cholangiographic examination revealed common bile duct dilation in all patients. Half of the patients had choledocholithiasis. Intrahepatic biliary dilation was present in 37.5% of the cases (Table 2).

Hepaticojejunostomy was the most common surgical procedure (62.5%). Two patients who underwent

Table 2. Cholangiographic findings of patients.

Variable	n
Common bile ductus dilation (>10 mm)	8
Common bile duct stone	4
Intrahepatic Bile Duct Dilation	3
Pneumobilia	2
Hook shaped CBD	2
Pancreatic duct visualization	2

CBD: Common bile duct

hepaticojejunostomy had not cholecystectomy before. Another common operation (25%) was choledochoduodenostomy. One of the patients underwent choledochoduodenostomy and gastroenterostomy in the same session due to the presence of a stenotic ulcer. Additionally, one patient underwent hepaticojejunostomy and gastroenterostomy in the same session. Case-based causes such as patient's age, comorbid factors, and less invasive techniques compared to hepaticojejunostomy were effective in the success of choledochodoudenostomy. With the increased experience of our clinic on choledochal anomalies, hepaticojejunostomy is applied more frequently only one of the cases in our study underwent portoenterostomy. In this case, dilation compatible with the choledochal cyst was detected during the operation which necessitated choledochal excision and portoenterostomy (Table 3).

The mean operative time was 165 min. in one patient, intraoperative iatrogenic small bowel injury developed and primary repair was performed and no complications developed. Postoperative anastomotic leakage developed in two patients and biliary leakage was resolved with medical treatment. Postoperative upper gastrointestinal bleeding developed in one patient which was controlled by endoscopic intervention. In one case, anastomotic leak was followed up medically, upper GIS bleeding occurred and surgical treatment was applied. During follow-up, the patient died of myocardial ınfarction (Table 3).

The mean postoperative hospital stay was 37 days. The causes of re-admission to the hospital within 90 days were wound infection (37.5%) and jaundice (12.5%). Any etiologic factor could not be detected by imaging and other methods in the patient who applied with jaundice. The mean follow-up period was 28 months (Table 3).

Table 3. Operative procedures, surgical morbidity, and clinical outcome.

	n
Surgicalapproach	
Hepaticojejunostomy	2
Hepaticojejunostomy with cholecystectomy	2
Hepaticojejunostomy with gastroenterostomy	1
Choledochoduodenostomy with cholecystectomy	1
Choledochoduodenostomy with gastroenterostomy	1
Portoenterostomy	1
Intraoperative Complication	
Iatrogenic small bowel injury	1
Postoperative complication	
Anastomosis leak	1
GIS bleeding	1
Anastomosis leak + GIS bleeding	1
Evisceration	1
Clavien-Dindo	
1	1
2	3
3	2
4	1
5	1
Reoperation	
GIS bleeding with anastomosis leakage	1
Evisceration	1
Postoperative 90-day mortality	1
90-day readmission	
Wound infection	2
Icterus	1

	Mean	Median	Range	Minumum-maximum
Mean operative time (min)	165	145	255	105-360
Postoperative Length of hospital stay (day)	37	28	82	8-90
Mean follow up (month)	28	26	47	6-53

GIS: Gastrointestinalsystem

DISCUSSION

The most important feature of the extrahepatic biliary system is the frequent occurrence of anatomic variations. The CBD typically enters the posteromedial aspect of the second portion of the duodenum. Sometimes the papilla of Vater may end in atypical anatomic localization, including the stomach, pyloric duct, duodenal bulb, and the third or fourth part of the duodenum. This very rarely seen condition is called ectopic biliary drainage [3]. Abnormal opening of the common bile duct to the duodenum has been reported in a limited number of cases. The incidence of common bile duct anomaly was found to be 0.43-1.05% in large series in the literature [1,2].

The etiology of ectopic biliary drainage is unknown and it is thought to be caused by abnormalities seen during embryological development.

In the literature, it is reported that 50-95% of cases with ectopic biliary drainage are seen in the male sex. This might bring to mind an etiologic relation with Y chromosome-induced embryonic abnormality [1,3,7,8]. In our series, male gender dominance was present in support of the findings in the literature.

Patients with anomalous choledochal openings become symptomatic in the 5th or 6th decades of life with symptoms such as cholangitis and upper gastrointestinal bleeding [1,2,8]. Although the age range of the patients was between 36-78 years in our series, the median age at diagnosis was 55 years which was consistent with the literature.

Biliary colic pain is the most common presenting symptom in ectopic common bile duct openings and this symptom is present in 95-100% of the patients during presentation [1,8]. In a study by A. Taş et al., presenting symptoms were determined to be biliary colic (100%), cholangitis (60%), acute renal failure (10%), previous bleeding (20%), and liver abscess (20%) [5]. The presenting symptoms in our series were abdominal pain (50%) and biliary colic (25%).

Ectopic opening of the CBD is commonly associated with biliary tract diseases. Gallstone, choledocholithiasis, obstructive jaundice, cholangitis, pancreatitis and peptic ulcer are complications of ectopic CBD openings [1,2,7,8]. Malfunction of the valve mechanism is blamed for the occurrence of these complications. The dysfunction of this mechanism may allow intestinal bacteria and gastric contents to enter the biliary system, causing recurrent attacks of cholangitis, transient obstruction and liver abscess [2,12]. In our series, one patient presented with cholangitis. Elevation of cholestatic enzymes was detected in 75% of the patients. Leukocytosis was present in 50% of the cases. Half of the patients in the study had a history of cholecystectomy due to cholelithiasis. Cholelithiasis was detected in 2 of 4 cases without cholecystectomy and choledocholithiasis was found in 62.5% of all cases.

In many series in the literature, it has been stated that the ectopic opening was localized mostly in the bulbus part of the duodenum [2,3,5,13]. In 75% of the cases in our series common biler duct opened in bulbus and in 25% of the case post-bulbar opening was detected .

Biliary system pathologies are frequently reported in patients with ectopic choledochal opening [1,2,5,8]. In their series, Taş et al. found common bile duct dilation in all of their patients. Deformed CBD, such as hook-shaped CBD, has been described as a predominant feature of ectopic orifice of papilla in the literature. The hook-shaped configuration may result from acute angulation of the CBD caused by premature drainage of bile into the bulb [8]. In the series of Lee et al, cholangiography showed this hook-shaped configuration of the distal CBD in all patients. They therefore argued that the hook-shaped configuration of the distal CBD should be considered as a characteristic finding for ectopic choledochal opening in patients without any history of abdominal surgery other than simple cholecystectomy [8]. In our series, all patients had common bile duct dilation and 37.5% of the patients had choledochal dilation accompanied by intrahepatic dilation. In 25% of our patients with ectopic common bile duct opening, hook shaped appearance was detected in the common bile duct. Although ERCP is widely used in the diagnosis of ectopic common bile duct opening, there are publications in the literature stating that endoscopic ultrasonography (EUS) will also be useful [14].

Coexistence of common bile duct anomaly and duodenal ulcer is a common condition. Easy passage of duodenal content into the biliary tract (duodenobiliary reflux) and reflux of the bile to the duodenum without sphincteric control can cause biliary tract diseases, duodenal ulcer and ulcer complications [1,2,5]. Diseases of duodenal mucosa (ulcers and deformation/stenosis) and biliary system diseases are closely related to the location and morphology of the ectopic opening [1,2,5,8]. In our series, 62.5% of the patients had duodenal ulcer and 2 patients had a history of duodenal ulcer surgery.

Treatment of ectopic biliary drainage is associated with biliary pathology and complications of peptic ulcer [3]. Most symptomatic patients are treated using surgical or endoscopic methods. If choledocholithiasis is accompanied by abnormal opening of the common bile duct into the stomach or duodenum, endoscopic treatment may fail and surgical intervention may be necessary. The intramural portion of the duct is not fully developed in these patients [8]. Therefore, there is a high risk of perforation and/or bleeding during endoscopic sphincterotomy, so endoscopic sphincterotomy is not recommended in these patients. There is no consensus regarding the treatment of asymptomatic ectopic choledochal openings [1,3,8].

In the study performed by Disibeyaz et al, endoscopic approach was the primary treatment method. Fifty-two percent of the patients with dilated CBD (with or without stone) were treated with endoscopic methods and 16% of them with surgical methods [1]. In the same study, surgical treatment was performed in patients with stenosis in the distal common bile duct or stones larger than 2 cm that could not be extracted by endoscopic intervention. Thirty-two percent of the patients with dilated CBD who were symptomatic but had not choledocholithiasis became asymptomatic with supportive medical treatment [1]. However, recurrent cholangitis episodes developed in 13-20% of patients that underwent endoscopic treatment.

Surgery is the main treatment modality in most of the case reports in the literature. Choledochoduodenostomy, choledochotomy with stone extraction and hepaticojejunostomy are frequently reported surgical methods. In the series of Lee et al., 72% of the patients were treated surgically and the most common surgical method was choledochoenterostomy [8]. In our series, patients underwent hepaticojejunostomy (n=5), portoenterostomy (n=1), and choledochoduodenostomy (n=2). In one patient, choledochoduodenostomy and gastroenterostomy procedure was performed due to stenosis resulting from duodenal ulcer. We performed gastroenterostomy on one of the patients who underwent hepaticojejunostomy due to ulcer-related stenosis. The main factors that determine the operation technique are anatomical location of the common bile duct opening, associated biliary pathologies, previous ulcer surgery and ulcer complications.

In one case, iatrogenic small bowel injury developed during bridectomy and primary repair was performed, and any postoperative complications were not observed. Postoperative anastomotic leakage developed in two, postoperative GIS bleeding in one and evisceration in one patient. The patient who had GIS bleeding was treated with the endoscopic method. Primary repair was performed on the patient who developed evisceration. One patient who developed anastomotic leakage was followed up medically and the fistula regressed in the follow-up. One patient who was followed up for anastomotic leakage developed massive ulcer bleeding. Since the bleeding could not be stopped by endoscopic procedures, the patient was operated on. In the operation, the bleeding ulcer was repaired with primary sutures, and gastroduodenal artery ligation and gastroenterostomy were performed. In the postoperative period, open abdomen was followed up for a long time. Although bile leakage and GIS hemorrhage were controlled during the follow-up, exitus developed due to myocardial infarction. In this case, we think that hemostatic therapies administered due to GIS bleeding had an effect on the development of myocardial ınfarction in the background of coronary heart disease. We think that recurrent inflammation of the common bile duct and consequent deterioration of wound healing are effective in postoperative anastomotic leakage. We estimate that bleeding secondary to duodenal ulcer developing in the postoperative period is due to the change in duodenal pH level resulting from applied bilioenteric diversion.

According to Clavien 2-3 classification used in the scoring of postoperative complications, our postoperative complication rate was 62.5%. The mean postoperative hospital stay was 37 days, and the main reason for the long hospital stay was anastomotic leakage, ulcer bleeding and wound infection. Wound infection and jaundice were the most common causes of unplanned re-admissions to the hospital within 90 days. The patient with jaundice was followed up medically and bilirubin levels returned to normal levels. Patients with wound infection were controlled with daily dressings. There were no deaths due to biliary system disease at a mean follow-up of 27.8 months. Myocardial infarction was the cause of mortality in one patient.

In conclusion, the ectopic opening of CBD into the duodenum is a rare anomaly that can cause incidental findings as well as pathologies in the biliary system and duodenal region. This anomaly should be kept in mind especially in patients with recurrent cholangitis, biliary colic, choledocholithiasis and / or recurrent duodenal ulcer. In addition to endoscopic methods, surgical treatment methods can be applied. Patients undergoing surgical treatment should be closely monitored for postoperative anastomotic leakage, bleeding arising from duodenal ulcer and sphincterotomy site. There are limited number of studies in the literature regarding surgical treatment of common bile duct anomalies. Prospective studies are needed to compare the long-term results of surgical and endoscopic treatment methods.

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