

# Retrospective analysis of cases with an ectopic opening of the common bile duct into duodenal bulb

Adnan Taş<sup>1,A</sup>, Banu Kara<sup>1,C</sup>, Sehmuz Ölmez<sup>1,B</sup>, Mehmet Suat Yalçın<sup>1,B</sup>,  
Nevin Akçaer Öztürk<sup>1,C</sup>, Bunyamin Saritas<sup>2,D</sup>

<sup>1</sup> Department of Gastroenterology, Adana Numune Research and Education Hospital, Turkey

<sup>2</sup> Department of Gastroenterology, Faculty of Medicine, Mersin University, Turkey

A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation;

D – writing the article; E – critical revision of the article; F – final approval of the article

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## Address for correspondence

Adnan Taş

E-mail: dradnantas@gmail.com

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## Abstract

**Background.** Ectopic opening of the common bile duct (EOCBD) is a very rare entity. It has been reported in the 3<sup>rd</sup> or 4<sup>th</sup> portion of the duodenum, pyloric canal, duodenal bulb, and the stomach.

**Objectives.** The aim of this study was to evaluate the clinical characteristics, laboratory values and imaging studies of patients with ECBD into the duodenal bulb retrospectively.

**Material and methods.** The files of patients who underwent endoscopic retrograde cholangiopancreatography (ERCP) between January 2003 and November 2015 were reviewed. The demographic data, presentations, abdominal ultrasonography, computed tomography (CT), magnetic resonance cholangiopancreatography (MRCP), and ERCP findings of patients with ECBD into the duodenal bulb were evaluated retrospectively.

**Results.** Ectopic openings of the CBD into the duodenal bulb were found in 20 out of 3270 patients who had undergone ERCP. Twenty patients (15 males and 5 females) with a median age of 59 (40–88) years were included in the study. Ectopic opening of the CBD into the duodenal bulb were found in 20 patients (0.61%). Laboratory test abnormalities included: hyperbilirubinemia in 20 (100%) patients, leukocytosis in 14 (70%) patients, and an elevated serum alkaline phosphatase and gamma-glutamyl transferase level in 20 (100%) patients. Indications for ERCP were CBD dilatation and extrahepatic cholestasis (n = 20), cholangitis (n = 12), only choledocholithiasis (n = 7), and acute pancreatitis (n = 2).

**Conclusions.** In patients with recurrent duodenal ulcers and/or apical stricture with accompanying CBD dilatation, extrahepatic cholestasis and cholangitis, ECBD into the duodenal bulb should be considered.

**Key words:** ectopic opening of the common bile duct in the duodenal bulb, duodenal ulcer, apical stricture, biliary disease

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## Introduction

Congenital anomalies of the common bile duct (CBD) are usually detected in childhood, but it might be undetected until adulthood. Congenital anomalies include accessory biliary ducts, aberrant cystic duct, bile duct cysts, double CBD, and ectopic opening of the CBD (EOCBD).<sup>1</sup> The etiology of EOCBD has been referred to errors in embryogenesis. Ectopic opening of the CBD is a very rare entity. However, it has been reported in the 3<sup>rd</sup> or 4<sup>th</sup> portion of the duodenum, pyloric canal, duodenal bulb, and the stomach.<sup>2–5</sup> The clinical and endoscopic features, and the significance of EOCBD are not sufficiently known. It is associated with benign biliary diseases, but usually manifests with recurrent duodenal ulcer, bulbus deformity with accompanying dilatation of the extra- and intrahepatic bile ducts with pneumobilia and stone.<sup>5</sup> Cases presented with gallbladder and stomach cancer with EOCBD into the duodenal bulb have also been reported.<sup>6–8</sup>

We retrospectively evaluated the clinical characteristics of patients with EOCBD into the duodenum (EOD), as well as their transabdominal ultrasonography (TUS), computerized tomography (CT), endoscopy, magnetic resonance cholangiopancreatography (MRCP) and endoscopic retrograde cholangiopancreatography (ERCP) findings of this rare anomaly.

## Material and methods

In this study, we retrospectively evaluated the files of patients who underwent ERCP in a time frame of January 2003–November 2015 in the Department of Gastroenterology of Adana Numune Training and Research Hospital (Turkey). Patients with EOD were found in our ERCP records. The demographic data, presentations, medical records, TUS, CT, endoscopy and ERCP findings of patients with EOD were evaluated retrospectively. The Ethics Committee of Adana Numune Training and Research Hospital approved the study.

An ectopic opening of the duodenum was defined as the failure to demonstrate the papilla in its original location in the 2<sup>nd</sup> part of the duodenum and papilla is observed as an orifice in the bulb which was proved to be CBD in cholangiogram either with MRCP or ERCP. The following ERCP findings were analyzed: the presence or absence of dilated bile ducts, configuration of the CBD, the presence of biliary stones.

Patients with infected bile duct stones typically presented with fever, abdominal pain and jaundice, and in severe cases – hypotension and mental confusion.<sup>9</sup> In patients with duodenal ulcer and apical stricture, gastric biopsies were taken from the antrum to exclude *Helicobacter pylori* (*H. pylori*) gastritis by histopathological examination. Eradication of *H. pylori* was performed with 14-day of tetracycline-lansoprazole-metronidazole-bismuth

subsalicylate containing regimen therapy. After *H. pylori* eradication, ERCP was repeated.

Statistical analyses were performed using PASW Statistics v. 18 (SPSS Inc., Chicago, USA).

## Results

Twenty patients diagnosed with EOD were included in the study. There were 15 men and 5 women with a median age of 59 (40–88) years. Out of 3,270 patients who had undergone ERCP, EOD was found only in 20 patients (0.61%).

All patients presented with episodic biliary pain (epigastric and/or right upper quadrant). Twelve patients (60%) had fever, abdominal pain and jaundice. Two patients had acute renal failure because of nausea and vomiting, as well as partial apical stricture. Four cases had previous bleeding episodes. Twelve patients were admitted to emergency department with severe suppurative cholangitis. Four out of 12 patients were in septic shock with multiple organ failure due to liver abscess shown on CT and they received prompt medical treatment. After their clinical condition improved, ERCP was performed. We found duodenal ulcer and partial apical stricture in 16 patients with endoscopy. Proton pump inhibitors were given to patients with duodenal ulcers and partial apical stricture. *Helicobacter pylori* infection was diagnosed through histologic examination in 4 patients and *H. pylori* eradication was done by 14-day tetracycline-lansoprazole-metronidazole-bismuth subsalicylate containing regimen therapies. Afterward, ERCP was performed. General characteristics, clinical and laboratory findings of the patients on admission to emergency department and applied treatments of the patients are presented in Table 1.

Laboratory test abnormalities included: hyperbilirubinemia in 20 (100%) patients, an elevated serum alkaline phosphatase and gamma-glutamyl transferase level in 20 (100%) patients, and leukocytosis in 14 (70%) patients.

Indications for ERCP were CBD dilatation and extrahepatic cholestasis (n = 20), cholangitis (n = 12), only choledocholithiasis (n = 7), acute pancreatitis (n = 2). A total of 44 ERCP sessions were performed in these patients.

There was a history of simple cholecystectomy in 8 (0.4%) patients (gallbladder stones). These patients did not have any problems after ERCP.

A major duodenal papilla was found in a slit-like opening in the duodenal bulb (2 patients). Various stages of ulceration, bulbar deformity and partial apical stricture were found in the duodenal bulb of 16 (80%) patients.

Transabdominal ultrasonography and MRCP findings were extrahepatic bile duct dilatation, with or without intrahepatic duct dilatation (20 patients). Transabdominal ultrasonography revealed choledocholithiasis in 6 patients, and MRCP revealed choledocholithiasis in 18 patients. Computed tomography revealed liver abscess in 4 patients.

**Table 1.** General properties, characteristics of patients admitted to emergency department and indications for ERCP

Patient characteristic	Number (%)
Men	15 (75)
Women	5 (25)
Median age [years]	59
ERCP	3270
EOPCD	20
Clinical characteristics	
Biliary pain	20 (100)
Cholangitis (fever, abdominal pain and jaundice)	12 (60)
Acute renal failure	2 (10)
Previous bleeding	4 (20)
Liver abscess	4 (20)
Apical stricture	16 (80)
Indications for ERCP	
CBD dilation and extrahepatic cholestasis	20 (100)
Cholangitis	12 (60)
Choledocholithiasis	7 (35)
Acute pancreatitis	2 (10)
Total number of ERCPs	44

ERCP – endoscopic retrograde cholangiopancreatography; EOPCD – ectopic opening of the common bile duct; CBD – common bile duct.

In the ERCP findings, the extrahepatic bile duct was dilated (CBD >10 mm diameter in patients with cholecystectomy, other patients with CBD >7 mm diameter) in 20 (100%) patients, with or without intrahepatic bile duct dilatation. Eighteen (90%) patients had choledocholithiasis; 7 patients had cholelithiasis.

Surgical treatment for bulbar stricture and biliary ectopic opening was performed in 14 patients. These patients did not have any problems after the operation. Patients who rejected surgical treatment underwent biliary balloon dilatation. The patients underwent follow-ups for a mean of 12 months. There were no other clinical problems that occurred during 12 months.

## Discussion

In our study, we found that the patients with EOCBD into the duodenal bulb might be undetected until adulthood and it may present with recurrent duodenal ulcer, apical stricture and severe biliary complications. We found the incidence of EOCBD into the duodenal bulb in patients who underwent ERCP as 0.61%.

The true incidence of patients with an EOCBD into the duodenal bulb is unknown. In a study by Lee et al., 18 out of 16,541 patients had an EOCBD into the duodenal bulb.<sup>4</sup> In 0.1% of patients who had undergone ERCP,

EOCBD into the duodenal bulb was diagnosed.<sup>4</sup> In a study by Sezgin et al., the diagnosis of EOCBD into various sites of the upper digestive tract was detected in 11 patients out of 1,040 patients who underwent ERCP (1.05%) and EOCBD into the duodenal bulb was observed only in 4 patients out of 1,040 patients who underwent ERCP (0.38%).<sup>2</sup> Disibeyaz et al. reported EOCBD into the duodenal bulb was found in 53 cases out of 12,158 patients who underwent ERCP (0.43%).<sup>10</sup> Saritas et al. reported the frequency of ectopic biliary drainage in 2% of patients who underwent ERCP (10 out of 400 ERCPs).<sup>11</sup> In the present study, EOCBD into the duodenal bulb was found in 20 patients out of 3,270 patients who underwent ERCP (0.61%).

Ectopic opening of the CBD into the duodenal bulb may be undetected until adulthood. In the study by Lee et al., the median age was 51 years, in the study by Sezgin et al., the median age was 59.2 years. In the study of Disibeyaz et al., the median age of the group was 55 years. Saritas et al. reported an EOCBD in patients with a median age of 54 years.<sup>2,4,10,11</sup> In our study, the median age was 51 years.

An ectopic opening of the CBD is usually associated with biliary tract illness.<sup>12</sup> In the study by Lee et al., 10 patients (56%) had bile duct stones, 15 patients (83%) had abnormal liver function tests, 11 patients (73%) had obstructive jaundice, and 7 patients (39%) had cholangitis.<sup>4</sup> Another study by Sezgin et al. reported choledocholithiasis (7 patients), acute pancreatitis, CBD dilatation, extrahepatic cholestasis, and cholangitis.<sup>2</sup> In the study by Disibeyaz et al., 25 patients (64.1%) had cholecystectomy. Two of them had acalculous cholecystitis. Recurrent cholangitis was evident in 10 patients (25.6%).<sup>10</sup> In our study, indications for ERCP were CBD dilatation and extrahepatic cholestasis (20 patients), cholangitis (12 patients), only choledocholithiasis (7 patients), and acute pancreatitis (2 patients).

An important feature in EOCBD is its association with duodenal ulcers. Lee et al. reported that 13 (72%) of the 18 patients had a history of duodenal ulcer.<sup>4</sup> Sezgin et al. reported that 2 out of 4 patients (50%) had duodenal ulcers and resultant apical stricture.<sup>2</sup> Recurrent duodenal ulcer was found in 24 (61.5%) patients and 4 subjects (20.5%) underwent gastric bypass operation due to gastric outlet obstruction related to peptic ulcer in the study by Disibeyaz et al.<sup>10</sup> In our study, we found duodenal ulcers and partial apical stricture in 16 patients with an endoscope. Bile reflux into the duodenal bulb causes ulcer formation and increases pH. In high pH, bile acids may induce gastric and duodenal bulb mucosal damage. Gastric metaplasia may develop in the mucosa of the duodenal bulb.<sup>5–13</sup>

In conclusion, the opening of the CBD into the duodenal bulb may be associated with biliary and duodenal diseases. Particularly, in patients with recurrent duodenal ulcers and/or apical stricture associated with CBD dilatation, extrahepatic cholestasis and cholangitis, EOCBD into duodenal bulb should be considered.

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