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Original Article

Rare and unique endoscopic retrograde cholangiopancreatography (ERCP) findings: insights from a 24-year single-center retrospective study

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ABSTRACT

Aim: Rare conditions causing biliary obstruction, such as Fasciola hepatica infestation, SUMP syndrome, Lemmel syndrome, Wirsung duct stones, Mirizzi syndrome, and surgical clip migration, are infrequently encountered during ERCP. This study aims to analyze the clinical and demographic characteristics, diagnostic challenges, and management of these uncommon conditions in a single-center cohort.

Method: This retrospective study included 6,700 patients aged 18 and older who underwent ERCP between 2000 and 2024. Patients with common bile duct stones, benign fibrosis, or malignancy were excluded, resulting in 28 cases with rare biliary pathologies. Data on clinical presentations, imaging findings, and treatment outcomes were analyzed.

Results: Among the 6,700 patients, the incidence of Fasciola hepatica infestation was 0.06% (n = 4), SUMP syndrome 0.75% (n = 5), Lemmel syndrome 0.06% (n = 4), Wirsung duct stones 0.044% (n = 3), Mirizzi syndrome 0.15% (n = 10), and surgical clip migration 0.03% (n = 2). Cholangitis was the most common clinical presentation across most conditions, with notable differences in total bilirubin levels, common bile duct diameters, and associated imaging findings. All cases were successfully managed with ERCP, and no complications were observed.

Conclusion: This study underscores the importance of recognizing rare biliary pathologies in patients presenting with atypical symptoms or a history of biliary surgery. Although these conditions represent a small percentage of patients undergoing ERCP, their timely diagnosis and management are crucial to preventing complications. For this reason, we are presenting these rare conditions collectively in the literature for the benefit of endoscopists.

Keywords: endoscopic retrograde cholangiopancreatography, fasciola hepatica, sump syndrome, lemmel syndrome, clips migration

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Introduction

Endoscopic Retrograde Cholangiopancreatography (ERCP) is a complex but essential procedure with a wide range of indications. ERCP was first found in the 1960s as a diagnostic tool but its therapeutic abilities were rapidly realized (1). The most common indications for ERCP are choledocholithiasis, cholangitis, acute biliary pancreatitis, and unidentified obstructions in the biliary tree (2).

Choledocholololithiasis, which presents with jaundice and abdominal pain caused by obstruction of the common bile duct (CBD), is also encountered with interesting diagnoses other than benign fibrotic lesions. Fascioliasis is a zoonotic infection caused by Fasciola hepatica and is a rare pathology observed in ERCP, and there are studies reported in the literature. Apart from this, Mirzzi syndrome observed due to cholelithiasis and clip migration observed after laparoscopic cholecystectomy are also interesting ERCP findings observed in the literature. Another interesting diagnosis observed in ERCP after surgery is SUMP syndrome, which can be observed after bile duct surgery. Pancreaticolithiasis is one of the interesting diagnoses that we can diagnose and treat in ERCP without jaundice and obstruction in the bile ducts. Although these cases are presented as single cases or case series in the literature (2-5), we present them as a whole in this study based on a single center experience. In our study, we aimed to highlight the interesting and less commonly seen cases of ERCP findings. With this review of our experiences of ERCP, we hope to raise the clinical suspicion of physicians and share our experiences.

Methods

This observational retrospective study included 6,700 patients aged 18 years and older who underwent ERCP between 2000 and 2024 at the Endoscopy Center of Istanbul Training and Research Hospital. The medical reports and imaging findings of these cases were retrospectively reviewed using the hospital's medical record system to identify pathologies within the CBD.

Patients diagnosed with CBD stones/sludge, fibrosis, or malignancy were excluded from the study. The remaining cases, comprising rare or less commonly reported diagnoses and their prevalence, were analyzed in the context of international literature. The study received approval from the Ethics Committee of Istanbul Training and Research Hospital (Approval No: 2011-KAEK-50, Decision Date: January 10, 2025).

Based on the inclusion and exclusion criteria, 28 patients with unusual ERCP findings were selected for further analysis. These findings included rare conditions such as fascioliasis, SUMP syndrome, Lemmel syndrome, Wirsung calculus, Mirizzi syndrome, and bile duct clip migration. For these selected cases, demographic and clinical characteristics were recorded, including the most common presenting complaints at hospital admission, mean age, gender distribution, mean diameter of the CBD prior to ERCP, presence of a gallbladder, presence of gallbladder calculi, and mean total bilirubin levels prior to ERCP. The data were compiled and subjected to descriptive analysis.

Results

Of the 6,700 ERCP cases reviewed, 88.7% (n = 5,943) were diagnosed with choledocholithiasis, 4.2% (n = 281) with malignancy, and 6.01% (n = 94) with benign fibrosis; these cases were excluded from the study (Table 1). A total of 28 patients with rare or unusual pathologies were included in the study and categorized into six groups: Fasciola hepatica obstruction (n = 4; 0.06%), SUMP syndrome (n = 5; 0.75%), Lemmel syndrome (n = 4; 0.06%), Wirsung duct stones (n = 3; 0.044%), Mirizzi syndrome (n = 10; 0.15%), and clip migration (n = 2; 0.03%) (Table 1).

Table 1. Findings and Distribution of ERCP Diagnoses

Finding	Number	Percentage (%)
Calculi-Sludge	5943	88.7
Malignancy	281	4.2
Benign Fibrosis/Obstructive Lesion	94	6.01
Fasciola Hepatica	4	0.06
SUMP Syndrome	5	0.75
Clip Migration	2	0.03
Lemmel Syndrome	4	0.06
Mirizzi Syndrome	10	0.15
Wirsung Stone	3	0.04
TOTAL	6700	100

Table 2. Demographic and Clinical Characteristics of Patients with Rare ERCP Diagnoses

Cases	Total size	Age (mean)	Male (%)	Female (%)	HS (most)	CBD (mean, mm)	GS	TB (mg/dl)
Fasciola Hepatica	4	42.5	25	75	Cholangitis	14	-	9.6
SUMP Syndrome	5	57.8	20	80	Cholangitis	22	operated	3.6
Lemmel Syndrome	4	70.75	25	75	Abdominal pain	12	+	5.9
Wirsung Stone	3	63.3	100	0	Abdominal pain	6.6	+	1.06
Mirizzi Syndrome	10	47.4	70	30	Jaundice	6.5	+	2.7
Clip Migration	2	42.5	100	0	Abdominal pain	10	operated	4.7

HS: The complaint in hospital admission, CBD: Common bile duct diameter, GS: Gallbladder stone, TB: Total Bilirubin

Fasciola Hepatica Obstruction

Four patients (0.06%) were diagnosed with Fasciola hepatica infestation. The group included three females and one male, with a mean age of 42.5 years. The most common clinical presentation was cholangitis. The mean total bilirubin level was 9.6 mg/dL, and the mean diameter of the CBD was 14 mm. Ultrasound imaging revealed mobile and linear echogenic findings in two cases, and parasites were visualized during ERCP in all four cases (Table 2).

SUMP Syndrome

Five patients (0.75%) were diagnosed with SUMP syndrome. The mean age of this group was 57.8 years, consisting of four females and one male. All patients had a history of cholecystectomy and choledochoduodenostomy, and the most common presentation was cholangitis. The mean total bilirubin level was 3.6 mg/dL, and the mean CBD diameter was 22 mm (Table 2).

Lemmel Syndrome

Four patients (0.06%) were identified with Lemmel syndrome, with a mean age of 70.75 years. Three of these patients were female. The most common presentation was right upper quadrant (RUQ) pain. The mean total bilirubin level was 5.9 mg/dL, and the mean CBD diameter was 12 mm. All patients had cholelithiasis detected on pre-ERCP ultrasound, and three were diagnosed via MRCP before ERCP (Table 2, Figure 1).

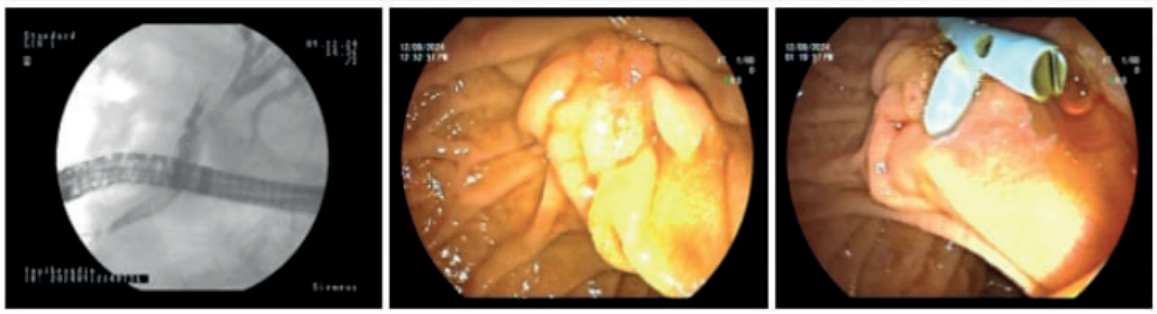

Figure 1. ERCP performed in a patient with Lemmel syndrome

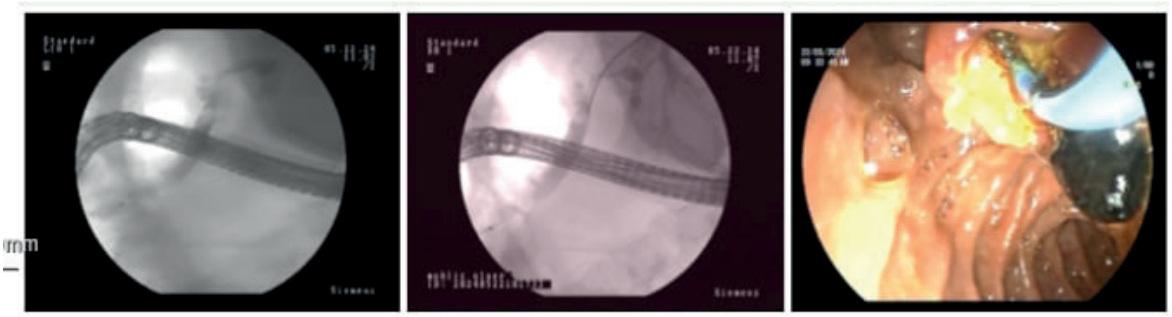

Figure 2. ERCP and stone extraction in a patient with Mirizzi syndrome

Wirsung Duct Stones

Three patients (0.044%) were found to have stones in the Wirsung duct, with a mean age of 63.3 years. All patients had a history of recurrent pancreatitis and presented with epigastric pain. The mean CBD diameter was 6.6 mm, and the mean total bilirubin level was 1.06 mg/dL. Pre-ERCP imaging (MRI and endoscopic ultrasound) revealed pancreatic duct dilation and pancreatic head stenosis, as well as cholelithiasis (Table 2).

Mirizzi Syndrome

Ten patients (0.15%) were diagnosed with Mirizzi syndrome, with a mean age of 47.4 years. Seven were male, and three were female. The most common clinical presentation was jaundice, with RUQ pain noted in half of the cases. Pre-ERCP imaging showed cholelithiasis in all cases, and one patient demonstrated pneumobilia. The mean total bilirubin level was 2.7 mg/dL, and the mean CBD diameter was 6.5 mm. Nine patients had type 1 Mirizzi syndrome, while one was classified as type 2 (Table 2, Figure 2).

Clip Migration

Two cases (0.03%) of clip migration were identified. Both patients were male, with a mean age of 42.5 years. The most common

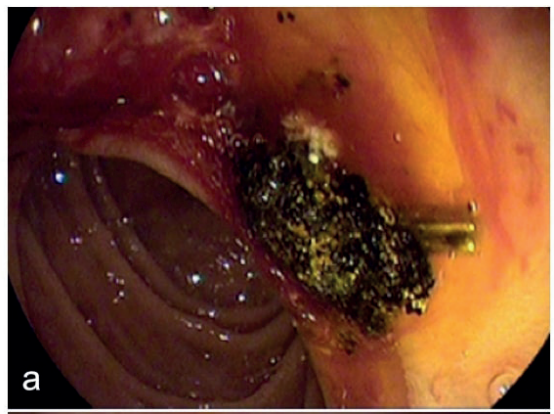

Figure 3. Stone extraction with ERCP in a patient with Mirizzi syndrome

presentation was RUQ pain. The mean CBD diameter was 10 mm, and the mean total bilirubin level was 4.7 mg/dL. One patient had undergone cholecystectomy 6 months prior, and the other 14 months prior. ERCP revealed bile duct sludge alongside migrated clips in both cases (Table 2, Figure 3).

Discussion

The findings of this study highlight the diversity and clinical significance of rare pathologies encountered during ERCP procedures. While the majority of cases involved common conditions such as choledocholithiasis (88.7%), malignancy (4.2%), and benign fibrosis (6.01%), this study focused on the less frequently observed diagnoses, comprising 0.42% (28 patients) of the total cohort. These included Fasciola hepatica obstruction, SUMP syndrome, Lemmel syndrome, Wirsung duct stones, Mirizzi syndrome, and clip migration. The rarity of these conditions underscores the importance of maintaining a high index of suspicion, especially in patients presenting with atypical symptoms or inconclusive imaging findings. By systematically analyzing these rare cases, this study contributes valuable insights into their clinical presentations, diagnostic challenges, and management strategies, providing a foundation for improved recognition and treatment in clinical practice.

Fasciola hepatica is a rare trematode infection in which the parasites can cause obstruction in the biliary tract. Diagnosing fascioliasis during the biliary phase can be challenging due to its wide variety of clinical presentations. In our study, 4 out of 6,700 patients (0.06%) were found to have obstruction of the CBD caused by F. hepatica. ERCP was performed, and cholangiography revealed filling defects in the CBD. Following sphincterotomy, adult trematode flukes were identified as the cause of obstruction. Encountering F. hepatica flukes during ERCP is rare. Our findings align with the case review by Sharma et al., (1) who also reported successful sphincterotomy and fluke extraction, followed by CBD stenting. In their case, anaphylaxis occurred—a known but serious complication of parasitic infections. Similarly, Aghajanzadeh et al. described a case where F. hepatica eggs obstructed the CBD, and the patient underwent sphincterotomy and stenting, ultimately recovering without significant complications (3,4).

In contrast, Rinaldi et al. reported a case of F. hepatica presenting as a peripancreatic mass. Their patient exhibited elevated bilirubin levels, with ERCP revealing CBD stenosis and fibrosis (5). In our study, cholangitis emerged as the most common clinical presentation, consistent with other reports. However, our patients exhibited a higher mean total bilirubin level (9.6 mg/dL) compared to similar cases in the literature. Additionally, one of our patients presented with a unique ultrasonographic finding of linear, mobile lesions within the gallbladder, distinct from gallstones. The mean CBD diameter in our cases was 14 mm, which exceeds the average range of 8–12 mm typically observed in choledocholithiasis cases (6). Given that fascioliasis is endemic to specific regions, we recommend that all patients presenting with biliary obstruction or cholangitis should be queried about their travel history to endemic areas (2).

Lemmel syndrome is a rare condition characterized by obstruction and jaundice caused by a periampullary duodenal diverticulum. ERCP is often the primary treatment modality for this condition. In our study, 4 out of 6,700 patients (0.06%) were diagnosed with Lemmel syndrome. A periampullary diverticulum can lead to obstruction through several mechanisms, including direct compression of the CBD or ampulla of Vater, dysfunction of the sphincter of Oddi, or chronic fibrosis of the papilla (7). Despite its rarity, Lemmel syndrome is a critical differential diagnosis for patients presenting with obstructive jaundice.

The diagnosis can be made using imaging techniques such as CT or MRCP, but the gold standard remains lateral-view endoscopy

during ERCP (8). Goroztieta-Rosales et al. reported a similar case of Lemmel syndrome in a patient presenting with RUQ pain and elevated bilirubin levels. The diagnosis was confirmed via ERCP (7). Similarly, Bernshteyn et al. described an ERCP case where impacted food was identified within the diverticulum and successfully removed using a snare (9). The incidence of duodenal diverticula is reported to be approximately 11% in patients older than 70 years, but the exact incidence of Lemmel syndrome remains unknown. In our cohort of 6,700 ERCP cases, the incidence of Lemmel syndrome was 0.06%. The mean age of patients in our study was 70.75 years, consistent with findings in the literature. The majority of patients presented with RUQ pain and a mean CBD diameter of 12 mm, both of which align with previously reported clinical characteristics of Lemmel syndrome (6). These findings underscore the importance of considering Lemmel syndrome in elderly patients presenting with biliary obstruction, especially when a duodenal diverticulum is identified on imaging.

Pancreatolithiasis can occur in the Wirsung or Santorini ducts, the side branches of pancreatic ducts, or within the pancreatic parenchyma. MRCP and EUS are highly sensitive diagnostic tools for detecting pancreatolithiasis (10-13). In our study, all three cases of pancreatolithiasis involved male patients with a history of recurrent pancreatitis. The mean age of these patients was 63.3 years, and two had significant histories of alcohol use. The incidence of pancreatolithiasis in our cohort was 0.044%, reflecting the rarity of this condition. Notably, the mean CBD diameter and total bilirubin levels were within normal ranges in these cases, which is consistent with the absence of significant biliary obstruction.

Mirizzi syndrome is a rare cause of obstructive jaundice, resulting from a stone impacted in the cystic duct or neck of the gallbladder (14). According to the literature, the incidence of Mirizzi syndrome requiring surgical intervention ranges from 0.07% to 1.4% (15). In our cohort of 6,700 ERCP cases, the incidence of Mirizzi syndrome was notably lower, at 0.15%. Among our cases, one was classified as type 2 Mirizzi syndrome, while the remaining cases were type 1. These findings underscore the rarity of Mirizzi syndrome in clinical practice and highlight the need for a high index of suspicion in patients presenting with obstructive jaundice and relevant imaging findings.

Surgical clip migration into the CBD is one of the rarest causes of jaundice, with fewer than 100 cases reported in the literature (16). The most widely accepted theory suggests that chronic inflammation near the CBD leads to erosion, particularly in cases where the cystic duct is anatomically close to the CBD. Migration has been reported to occur up to 14 years post-cholecystectomy, although the majority of cases are documented within 2 years of surgery (17,18). Photi et al. reported a case of clip migration 16 years post-surgery, successfully managed with ERCP, similar to the cases in our study (19). In our series, all patients presented with RUQ pain and had a mean total bilirubin level of 4.7 mg/dL, approximately three times the upper normal limit. All cases were successfully treated with ERCP without any complications, highlighting the effectiveness of this approach in managing this rare condition.

SUMP syndrome is an exceptionally rare condition, primarily occurring in patients with a history of side-to-side choledochoduodenostomy (CDD) or open cholecystectomy.In SUMP syndrome, the portion of the CBD distal to the CDD becomes a reservoir for debris due to poor drainage, forming what is referred to as a “sump” (20,21). Abbarh et al., in their case series, reported three patients with SUMP syndrome, all with a history of CDD performed during open cholecystectomy. Two of their patients were successfully treated with a combination of percutaneous transhepatic cholangiography (PTC) and ERCP, while one patient did not return for the planned ERCP (21). In our study, both patients diagnosed with SUMP syndrome were successfully managed with ERCP alone, without the need for PTC. Similarly, Abraham

et al. described a patient with a history of CDD who presented with cholangitis and was treated with urgent ERCP (20).

Granata et al. reported a unique approach where they utilized an endoscopic overstitch device to reduce the diameter of the CDD, thereby preventing further accumulation of debris in the sump (22). In our study, cholangitis was the most common clinical presentation among patients with SUMP syndrome. The mean CBD diameter in this group was 22 mm, the largest observed in our study, reflecting the significant drainage dysfunction associated with this condition. All patients had a history of CDD, consistent with findings by Ağca et al., who reported SUMP syndrome in four cases of CDD and one case of choledochojejunostomy (23). These observations reinforce the association between prior biliary surgeries and the development of SUMP syndrome, emphasizing the importance of tailored treatment strategies in such cases.

The primary limitation of this study is its retrospective design, which inherently depends on the accuracy and completeness of recorded data. Additionally, the rarity of the conditions studied resulted in a small sample size for each subgroup, limiting the generalizability of the findings. The study was conducted at a single center, which may not reflect variations in patient demographics or clinical practices across different regions. Finally, the lack of long-term follow-up data for the treated patients precludes an analysis of recurrence rates or long-term outcomes.

Conclusion

This study highlights the importance of recognizing rare and unusual causes of biliary obstruction, such as Fasciola hepatica infestation, SUMP syndrome, Lemmel syndrome, Wirsung duct stones, Mirizzi syndrome, and surgical clip migration. Although these conditions represent a small percentage of patients undergoing ERCP, their timely diagnosis and management are crucial to preventing complications. Advanced imaging techniques, such as MRCP and EUS, alongside ERCP, play a vital role in the accurate diagnosis and treatment of these rare pathologies. Our findings emphasize the need for a high index of suspicion, particularly in patients with atypical presentations or relevant surgical histories. Future studies with larger cohorts and prospective designs are needed to further explore these rare conditions, improve diagnostic accuracy, and optimize treatment strategies.

Ethical approval

The study was performed according to the Helsinki Declaration. The study protocol was approval by the Ethics Committee of Istanbul Training and Research Hospital (Approval No: 2011-KAEK-50, Decision Date: January 10, 2025).

Author contribution

The authors confirm contribution to the paper as follows: Study conception and design: EÇ, OOK, MMS; data collection: EÇ, OOK, SZK, MAD, BD; analysis and interpretation of results: EÇ, OOK, SZK, MAD, BD; draft manuscript preparation: EÇ, OOK, MMS. All authors reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The authors declare that there is no conflict of interest.

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