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Refractory Choledocholithiasis Causing Endogenous Endophthalmitis: A Case Report

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Male, 73-year-old
Final Diagnosis: Cholangitis • choledocholithiasis • endogenous endophthalmitis
Symptoms: Fever • visual acuity loss
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology • Surgery

Objective: Rare disease

Background: Endogenous bacterial endophthalmitis is caused by a breach of the blood-ocular barrier by pathogens originating from distant infective foci. Here, we report a case of endogenous endophthalmitis due to cholangitis complicated by common bile duct stones, which is a rare source of infection.

Case Report: A 73-year-old man with type II diabetes mellitus underwent endoscopic choledocholithotripsy 20 years ago and laparoscopic cholecystectomy 18 years ago. He had choledocholith-related cholangitis 6, 5, and 1 years previously and 4 times in the last year and underwent endoscopic choledocholithotripsy each time. Three days after the last surgery, the patient developed right endogenous endophthalmitis and vitrectomy was performed. Four months later, the patient relapsed with cholangitis and required surgery for recurrent endophthalmitis. Roux-en-Y choledochojejunostomy was performed with curative intent, and the patient was followed up for 5 years without recurrence of choledocholith, cholangitis, or endophthalmitis.

Conclusions: The recommended treatment strategy for patients diagnosed with common bile duct stones or choledocholithiasis is stone extraction. Endoscopic retrograde cholangiopancreatography (ERCP) with endoscopic intervention is a widely accepted procedure. However, in cases of recurrent choledocholithiasis, the rate of recurrence increases and the interval between ERCP becomes shorter in proportion to the number of recurrences. In such intractable cases requiring numerous sessions of endoscopic stone removal, bypass Roux-en-Y choledochojejunostomy should be performed to prevent possible rare complications such as endogenous bacterial endophthalmitis.

Keywords: Cholangitis • Choledochostomy • Endophthalmitis • Endoscopy, Digestive System

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/938890

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Background

Endophthalmitis is a rare but potentially sight-threatening disease characterized by marked inflammation of intraocular spaces. This ocular pathology can be classified as exogenous or endogenous endophthalmitis, depending on the route of infection. Endogenous bacterial endophthalmitis, also called metastatic bacterial endophthalmitis, is caused by a breach of the blood-ocular barrier by pathogens originating from distant infective foci, such as endocarditis, liver abscess, and urinary tract infection. Endogenous bacterial endophthalmitis is generally rarer than exogenous endophthalmitis, accounting for 2% to 8% of all cases [1]. However, recent research revealed the prevalence to be 26.4% [2] or 41% [3], indicating the involvement of geographic, genetic, or alimentary factors. The most frequent source of infection is endocarditis, followed by the gastrointestinal tract and genitourinary tract in North America and Europe [1]; however, liver abscess is the most frequent source in East Asia [4]. There is no age or sex predilection in the incidence of endogenous endophthalmitis [2]. According to a worldwide systemic survey of endogenous bacterial endophthalmitis, diabetes was the most common predisposing medical condition [5]. Herein, we report the first case of endogenous endophthalmitis due to cholangitis complicated by refractory common bile duct stones, which is a rare infection source.

Case Report

A 73-year-old man with a history of type II diabetes mellitus presented with a fever of 40°C and abdominal pain and was referred to our hospital. He had undergone endoscopic removal of common bile duct stones 20 years ago and laparoscopic cholecystectomy for gallbladder stones 18 years ago. He had choledocholith-related cholangitis 6, 5, and 1 year previously and underwent endoscopic choledocholithotripsy each time. Additionally, choledocholith-related cholangitis recurred 3 times in the last year, and endoscopic choledocholithotripsy was performed each time. One month after the last endoscopic choledocholithotripsy, the patient was referred to our hospital for epigastralgia. Laboratory findings on admission showed: white blood cell count 14 260/μL, serum C-reactive protein 11.6 mg/dL, total bilirubin 3.1 mg/dL, aspartate aminotransferase 46 U/L (normal range 8-38 U/L), alanine aminotransferase 71 U/L (4-44 U/L), alkaline phosphatase 458 U/L (104-338 U/L), and gamma-glutamyl transpeptidase level 369 U/L (16-73 U/L). Computed tomography on admission revealed common bile duct stones; therefore, endoscopic choledocholithotripsy and biliary drainage were performed. *Klebsiella pneumoniae* and *Escherichia coli* were detected in blood and bile cultures. Cefmetazole was administered at a dose of 3 g/d. Three days after admission, the patient's general status improved, but he experienced impaired vision and sensation of something rolling around his right eye for the first time. The patient was diagnosed with endogenous bacterial endophthalmitis of the right eye (**Figure 1**), for which anterior chamber irrigation, lensectomy, and vitrectomy were performed. *Klebsiella pneumoniae* was detected in the aqueous humor of the right eye. Visual acuity improved after surgery. However, 4 months after the operation, choledocholith-related cholangitis recurred (**Figure 2**), and endoscopic choledocholithotomy was performed. The vision in the right eye started to deteriorate 5 days postoperatively owing to the recurrence of endophthalmitis. Hence, vitrectomy was performed 3 times and the final vision was maintained at 0.3. Finally, a Roux-en-Y choledochojejunostomy was performed for refractory choledocholith-related cholangitis. The postoperative course was good, and the patient was followed up for 5 years without recurrence of choledocholith, cholangitis, or endophthalmitis.

Figure 1. In the right eye, fundoscopy shows diffuse opacity of vitreous body and cannot show ocular fundus by decreased permeability (**A**, right eye; **B**, left eye).

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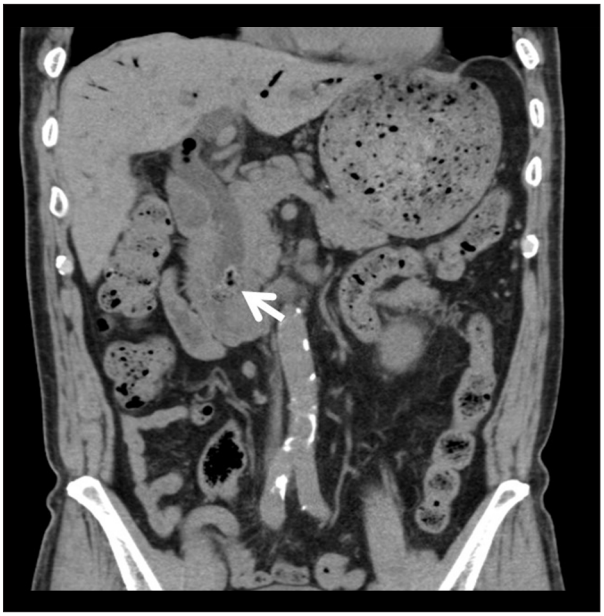

Figure 2. Abdominal CT reveals a mass of diameter 24 mm, which is suspected to be choledocholith (arrow), in the duodenal end of the common bile duct.

Discussion

The majority of cases of endogenous bacterial endophthalmitis have comorbidities, such as diabetes mellitus, and tumor, or autoimmune diseases, and were immunocompromised owing to immunosuppressant drug therapy [1,4-6]. Diabetes is the most common comorbidity, with an incidence of 29% to 61.9% [6]. Moreover, our patient also had diabetes mellitus. There is no age or sex predilection in the incidence of endogenous endophthalmitis [2].

The most frequent source of infection differs across countries; however, liver abscesses are the most common cause of hepatobiliary infections. There has been only 1 case of endogenous endophthalmitis with a source of cholangitis without cholecystitis [7] and the causative organism was *Pseudomonas aeruginosa*. Cholangitis generally occurs acutely, with evident clinical manifestations, allowing prompt diagnosis and appropriate treatment. Therefore, endogenous endophthalmitis may no longer occur. In our case, intractable cholangitis caused by refractory common bile duct stones treated with repeated endoscopic lithotripsy induced endogenous endophthalmitis due to continuous biliary infection.

Here, the causative organism was *Klebsiella pneumoniae*. Liver abscesses in patients affected with *Klebsiella pneumoniae* cause extrahepatic complications resulting from bacteremic dissemination, including endophthalmitis, meningitis, necrotizing fasciitis, and other illnesses, called invasive liver abscess syndrome [8]. The invasive nature of some *Klebsiella pneumoniae* strains includes a hypermucoviscous phenotype associated with serotypes K1 and K2 and the regulator of mucoid phenotype A (rmpA) gene. The prognosis of patients with endophthalmitis caused by *Klebsiella pneumoniae* is very poor, with more than 85% of patients having severe visual deficits [4,9,10]. Early diagnosis and appropriate treatment are necessary for a better prognosis with visual recovery. In our case, repeated vitrectomy until the subsiding of the inflammation played a vital role in treating endogenous bacterial endophthalmitis.

It is recommended that patients diagnosed with common bile duct stones or choledocholithiasis undergo stone extraction [11]. Endoscopic retrograde cholangiopancreatography (ERCP) with endoscopic intervention has been widely accepted for choledocholithiasis owing to its recent development. However, a minority of patients experienced recurrent choledocholithiasis and repeat ERCP had choledochal complications at a relatively high rate [12]. The rate of subsequent recurrence increased, and the interval between ERCP became shorter in proportion to the number of recurrences, as in our case. Multiple surgical treatments aggravate the Oddi sphincter injury, and subsequent reflux biliary infection and bile stasis may play an important role in the pathogenesis of recurrent stones [12,13]. In intractable cases requiring numerous endoscopic stone removal sessions, bypass Roux-en-Y choledochojejunostomy can be a definitive treatment [13].

Conclusions

Endogenous endophthalmitis should be considered for intractable common bile duct stones with *Klebsiella pneumoniae* infection, and bypass choledochojejunostomy should be considered at an appropriate time.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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