

Recurrent cholangitis caused by a giant duodenal diverticulum (Lemmel syndrome) – effective treatment with endoscopic sphincterotomy

I. Mendes^{1,2}, F. Vara-Luiz^{1,2}, G. Nunes^{1,2}, J. Cruz³, J. Veloso¹, J. Fonseca^{1,2}

(1) Gastroenterology Department, Hospital Garcia de Orta, Almada, Portugal; (2) Egas Moniz Center for Interdisciplinary Research (CiiEM), Egas Moniz School of Health and Science, Almada, Portugal. PaMNEC – Grupo de Patologia Médica, Nutrição e Estudos Clínicos; (3) Radiology Department, Hospital Garcia de Orta, Almada, Portugal..

To the Editor,

A 70-year-old male was admitted with abdominal pain and jaundice. Past medical history was relevant for severe chronic obstructive pulmonary disease and multiple cardiovascular risk factors. Biological tests revealed leukocytosis ($18.5 \times 10^9/L$), hyperbilirubinemia (total bilirubin of 4.9 mg/dL and direct bilirubin of 2.8 mg/dL) and elevated C reactive protein (13.64 mg/dL) and liver enzymes (alanine aminotransferase of 562 UI/L, aspartate aminotransferase of 689 UI/L and alkaline phosphatase of 378 U/L). Abdominal CT showed a dilated biliary tree and no obstructive lesions. The patient started on antibiotics due to suspected acute cholangitis. A magnetic resonance cholangiopancreatography was performed, showing a giant duodenal diverticulum (shown in Fig. 1) involving the distal segment of the common bile duct almost circumferentially, leading to its collapse and upstream dilatation of intra and extrahepatic bile ducts, without luminal stones or stenosis. Imaging findings were consistent with Lemmel syndrome (LS).

Giving the expectable favorable course, a conservative ambulatory follow-up was planned and the patient

a surgical approach. Endoscopic assessment revealed the major duodenal papilla within a large diverticulum (shown in Fig. 2a,b). Dilatation of intra and extrahepatic bile ducts with narrowing of the distal portion of the common bile duct in relation with the diverticulum was observed in cholangiography. A complete contrast filling of the duodenal diverticulum was possible to obtain (shown in Fig. 2c,d). Biliary endoscopic sphincterotomy (ES) was successfully performed. The patient was discharged after 7 days of antibiotics, remaining free of cholangitis episodes during follow-up.

Most duodenal diverticula are asymptomatic and do not require treatment, being accidentally diagnosed by cross-sectional imaging or upper GI endoscopy. LS is a rare entity defined as obstructive jaundice caused by a periampullary duodenal diverticulum. Pathophysiology includes chronic fibrosis secondary to inflammation (diverticulitis) leading to obstruction, sphincter of Oddi dysfunction, or direct external compression of the common bile duct by the diverticulum and its content (1,2). Patients with LS can present with complications such as acute pancreatitis or cholangitis, which can be recurrent if the underlying cause is left untreated.

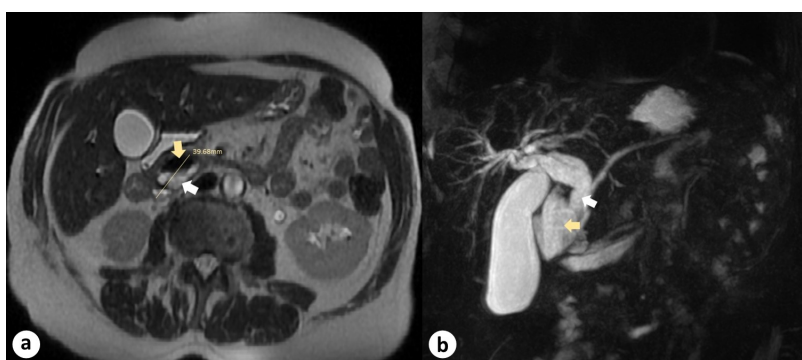


Figure 1. — a,b: Magnetic resonance cholangiopancreatography showing a duodenal diverticulum (yellow arrow) with almost 4cm involving the distal segment of the common bile duct (white arrow).

discharged after 7 days of antibiotics. Nevertheless, he was readmitted with a second episode of acute cholangitis 8 months later. Considering the advanced age and comorbidities, treatment through endoscopic retrograde cholangiopancreatography (ERCP) was preferred over

Correspondence to: Ivo Mendes, Avenida Torrado da Silva, 2805-267 Almada, Portugal. Phone: 00351919932369. Email: ivoivomendes@gmail.com

Submission date: 29/01/2024
Acceptance date: 09/02/2024

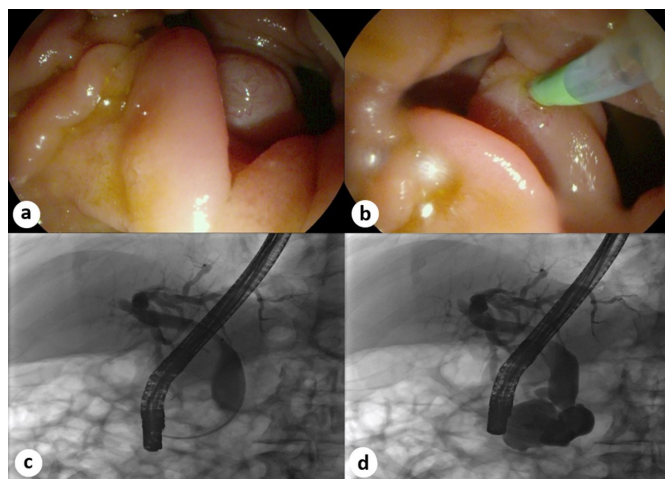


Figure 2. — a: Endoscopic view of the major duodenal papilla within a large diverticulum; b: Biliary cannulation; c: Cholangiography revealing dilatation of intra and extrahepatic bile ducts with narrowing of the distal portion of the common bile duct; d: Complete contrast filling of the duodenal diverticulum.

Therapeutic options for LS include surgery (mainly diverticulectomy), endoscopic intervention and conservative management (2,3). However, there is no standardized approach. Treatment should be individualized according to the patient characteristics and preferences, as well as the experience of the medical team (4).

The authors present a case of LS that was successfully treated with ES. Despite the need for further research, some case reports suggest that an endoscopic intervention, either by sphincterotomy, biliary stenting or simply diverticular lavage, may represent an effective and minimally invasive therapeutic option to treat and prevent biliopancreatic complications, especially in high-risk LS patients who are poor surgical candidates (3,5).

Statement of Ethics

The patient has given informed consent for publication (including the publication of images).

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Data Availability Statement

The complete data of this manuscript are not publicly available due to the patient's privacy but are available from the corresponding author upon reasonable request.

References

1. GOROZTIETA-ROSALES LM, GÓMEZ-FARÍAS J, LÓPEZ-GARCÍA KD, DAVILA-RODRIGUEZ DO. Lemmel syndrome: an extraordinary cause of obstructive jaundice-a case report. *J Surg Case Rep.* 2022(1):1-3.
2. BERNSHTEYN M, RAO S, SHARMA A, MASOOD U, MANOCHA D. Lemmel's Syndrome: Usual Presentation of an Unusual Diagnosis. *Cureus.* 2020; 12(4):1-6.
3. LOVE JS, YELLEN M, MELITAS C, YAZICI C, ZAR F. Diagnosis and Management of Lemmel Syndrome: An Unusual Presentation and Literature Review. *Case Rep Gastroenterol.* 2022; 16(3):663-74.
4. AGÚNDEZ MC, GUERRA DL, PÉREZ JF, FERNÁNDEZ GB. Síndrome de Lemmel: ictericia obstructiva secundaria a divertículo duodenal. *Cir Esp.* 2017;95(9):550-1.
5. CHIANG TH, LEEYC, CHIU HM, HUANG SP, LIN JT, WANG HP. Endoscopic therapeutics for patients with cholangitis caused by the juxtapapillary duodenal diverticulum. *Hepatogastroenterology.* 2006;53(70):501-5.