СЛУЧАЙ ИЗ ПРАКТИКИ

INTERMITTENT OBSTRUCTIVE JAUNDICE DUE TO A DUODENAL DIVERTICULUM: LEMMEL'S SYNDROME INITIALLY MISSED ON CT BUT LATER EVINCED WITH MAGNETIC RESONANCE CHOLANGIOGRAPHY AND ENDOSCOPY

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Due to a mechanical obstruction of the common bile duct, Lemmel's syndrome arises when a duodenal diverticulum induces obstructive jaundice. Intermittent obstructive jaundice is a rare conditionreported in 2-7% of patients undergoing studies. Although the majority of the diverticula are asymptomatic when the symptoms are present, they usually consist of jaundice, abdominal pain, or acute cholangitis; these symptoms can be intermittent. The imaging methods in diagnosing Lemmel's syndrome are crucial; the CT scan and magnetic resonance cholangiography (MRC) revealed periampullary diverticula on the medial wall of the second portion of the duodenum. Treatment varies depending on the aetiology and underlying cause. However, diverticulectomy remains the standard of care.

This report describes a 75-year-old male who attended the emergency department with jaundice, acute abdominal pain, and a weight loss history. Through laboratory studies and duodenal endoscopy, obstructive jaundice due to Lemmel's syndrome was evinced and received appropriate treatment. Lemmel's syndrome was initially missed in the CT evaluation but identified in the MRC and endoscopic view of duodenal endoscopy. Clinicians should always consider diverticulum as a possible cause of pancreatitis and obstructive jaundice. Readers will find an updated review of the literature.

Keywords: Lemmel's syndrome; obstructive jaundice; magnetic resonance cholangiography; endoscopy.

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РЕЦИДИВИРУЮЩАЯ МЕХАНИЧЕСКАЯ ЖЕЛТУХА ВСЛЕДСТВИИ ДИВЕРТИКУЛА ДВЕНАДЦАТИПЕРСТНОЙ КИШКИ: СИНДРОМ ЛЕММЕЛЯ, ПРОПУЩЕННЫЙ ПРИ МСКТ, ПОЗЖЕ ВЫЯВЛЕННЫЙ ПРИ МАГНИТНО-РЕЗОНАНСНОЙ ХОЛАНГИОГРАФИИ И ЭНДОСКОПИИ

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Синдром Леммеля возникает из-за механической обструкции общего желчного протока, когда дивертикул двенадцатиперстной кишки вызывает механическую желтуху. Рецидивирующая механическая желтуха является редким состоянием, которое встречается у 2-7% пациентов при обследовании. Большинство дивертикулов протекают бессимптомно, но когда симптомы все же присутствуют, это обычно проявляется желтухой, болью в животе или острым холангитом; эти симптомы могут быть рецидивирующими. Методы визуализации в диагностике синдрома Леммеля имеют решающее значение. При помощи МСКТ и МР-холангиографии могут быть выявлены периампулярные дивертикулы на медиальной стенке второго отдела двенадцатиперстной кишки. Тактика лечения зависит от этиологии и сопутствующих факторов, однако стандартом лечения остается дивертикулэктомия.

В данном клиническом случае представлено наблюдение мужчины, 75 лет, обратившегося в отделение неотложной помощи с желтухой, острой болью в животе и потерей веса в анамнезе. При помощи лабораторных исследований и эндоскопии двенадцатиперстной кишки была выявлена механическая желтуха, вызванная синдромом Леммеля и было проведено соответствующее лечение. Первоначально синдром Леммеля был пропущен при МСКТ, но был выявлен при МР-холангиографии и эндоскопическом исследовании двенадцатиперстной кишки. Дивертикулы двенадцатиперстной кишки всегда должны рассматриваться как возможная причина панкреатита и механической желтухи. Статья дополнена обзором литературы.

Ключевые слова: синдром Леммеля, механическая желтуха, магнитно-резонансная холангиография, эндоскопия.

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Introduction.

Lemmel's syndrome is a rare condition, initially described in 1934 as aperiampullary duodenal diverticulum causing obstructive jaundice in the absence of gallstones [1]. The prevalence of duodenal diverticula ranges from 5-10%, increasing with age; 75% are of periampullary origin, and only 1% of these diverticula will trigger symptoms, being an incidental finding when studying another disease or with a routine endoscopy [2].

The symptomatic duodenal diverticulum is associated with increased incidence and presentation of biliopancreatic disease, secondary to extrinsic compression of the diverticulum to the bile duct [3]. Diverticulitis, bleeding, perforation, and fistula formation are examples of non-pancreaticobiliary consequences. Recurrent gallbladder or bile duct stones, obstructive jaundice, cholangitis, or severe pancreatitis are all examples of pancreaticobiliary problems [4].

Imaging studies arecritical to identify and diagnose Lemmel's syndrome, and awareness of this condition may prevent mismanagement [5]. An inexperienced clinician could identify this entity as diverticula filled with fluid and misdiagnose it as a pancreatic abscess, cystic neoplasm in the pancreatic head, or a metastatic lymph node. Many of these findings will be incidental, so we must consider a differential diagnosis of pseudocysts, pancreatic tumours, or metastasis [5].

This report describes a 75-year-old male who attended the emergency department with jaundice, acute abdominal pain, and weight loss history. Lemmel's syndrome was initially missed in the computed tomography (CT) performed at admission but later identified during magnetic resonance cholangiography (MRC) evaluation duodenal endoscopy. The reader will find an updated review of the literature.

Case presentation

A 75-year-old male attended the emergency department with a four months history of moderate abdominal pain predominantly in the right upper quadrant with radiation to the epigastrium accompanied by jaundice, nausea, and up to three times per day vomiting of intestinal content; the last months before admission, he had noticed weight loss, fullness, and early satiety with food intake.

Clinical history revealed a family history of Type 2 Diabetes Mellitus. Non-pathological history included poor diet in quantity and quality; the patient denied smoking and alcoholism.

Past medical history including a tonsillectomy in childhood, open cholecystectomy 16 years before due to uncomplicated acute lithiasis and cholecystitis; transurethral resection of the prostate due to prostate cancer not specified, six years before admission currently under vigilance. An abdominal ultrasound performed one week before showed dilation of the bile duct.

An abdominal CT and magnetic resonance cholangiography (MRC) performed on admission showed dilation of the common bile

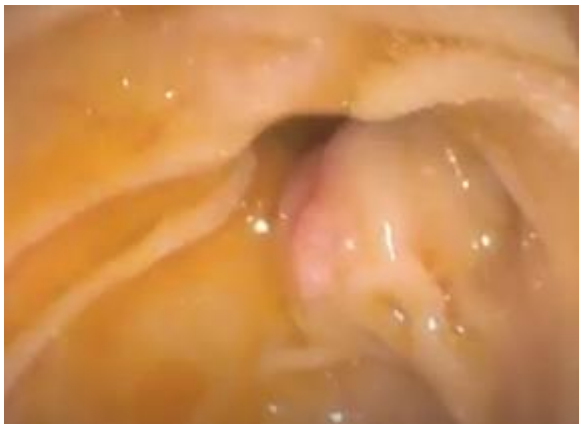


Fig. 1 a (Рис. 1 a)

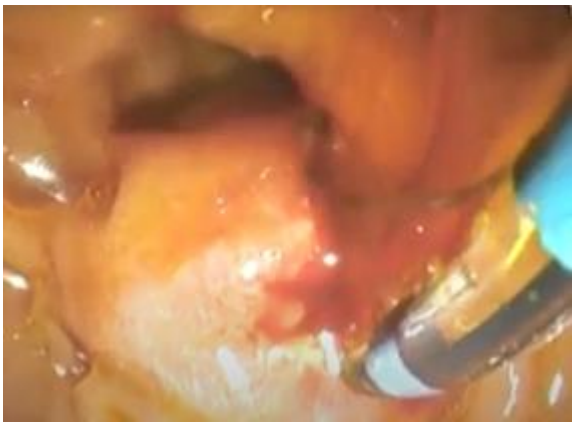


Fig. 1 б (Рис. 1 b)

Fig. 1. Endoscopic images.

A – periampullary duodenal diverticula. B – papilla cannulated with sphincterotome during sphincterotomy procedure.

Рис. 1. Эндоскопические изображения.

А – периампулярные дивертикулы двенадцатиперстной кишки. Б – сосочек, канюлированный сфинктеротомом во время процедуры сфинктеротомии.

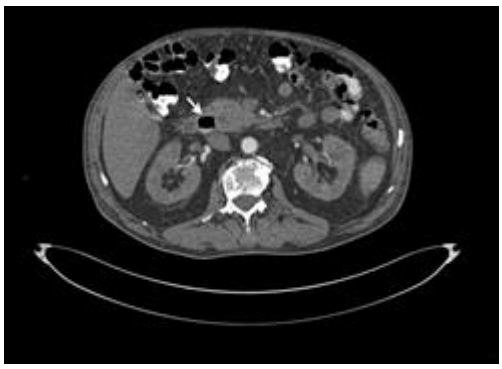


Fig. 2 a (Рис. 2 а)

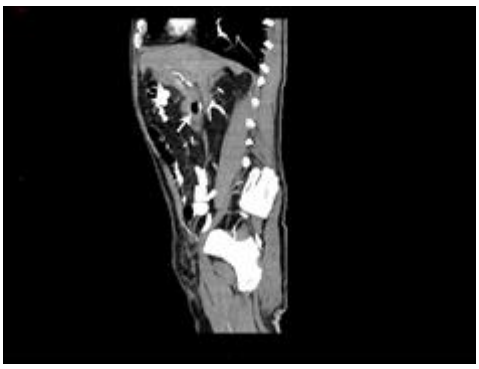


Fig. 2 b (Рис. 2 б)

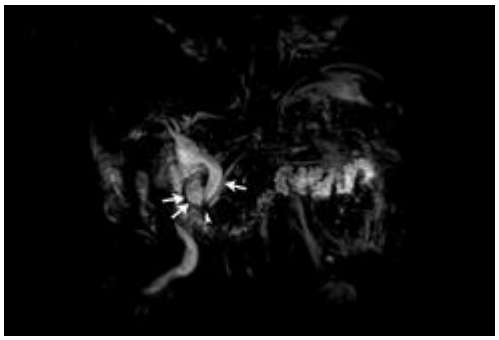


Fig. 2 c (Рис. 2 в)

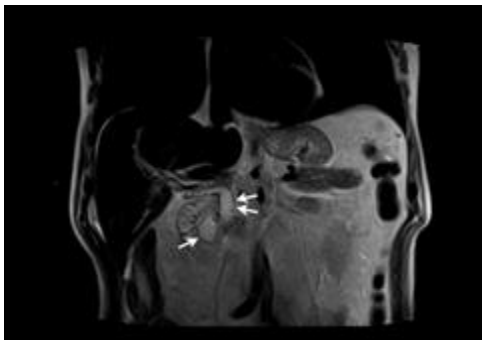


Fig. 2 d (Рис. 2 г)

Fig. 2. A, B – CT, abdomen, axial and sagittal planes, with double contrast, show a saccular image with air-fluid level dependent on the duodenum wall (white arrows) suggestive of a diverticulum. C – Magnetic cholangioresonance shows an increase in the calibre of the bile duct (white arrows) with a punctate termination was observed (white arrowhead), the content of iso-hypointense material suggestive of bile sludge; additionally, an image of a saccular aspect dependent on the duodenal wall is observed, with hypointense content in contact with the choledocus; and intrahepatic duct dilatation (double white arrows). D – T2 sequence of MRI in coronal plane shows the saccular image (white arrow) adjacent to the dilated bile duct (double white arrows).

Рис. 2. A, B – МСКТ органов брюшной полости, с двойным контрастированием, аксиальная и сагиттальная плоскости соответственно; визуализируется мешковидное образование с уровнем воздуха и жидкости, связанное со стенкой двенадцатиперстной кишки (белая стрелка) – дивертикул. C – МР-холангиография, увеличение калибра желчных протоков (белые стрелки) с точечным окончанием (остриё стрелки), содержание изогипоинтенсивного материала указывает на желчный сладж; дополнительно визуализируется мешотчатое образование в области стенки двенадцатиперстной кишки с гипоинтенсивным содержимым, контактирующим с холедохом; расширение внутрипеченочных протоков (две белые стрелки). D – МРТ, Т2-ВИ, корональная плоскость; визуализируется мешотчатое образование (белая стрелка), примыкающее к расширенному желчному протоку (две белые стрелки).

duct without alteration in the pancreas. Laboratory studies showed a cholestatic pattern and CA 19-9 nega-tive. Due to the inconclusive CT findings, an MRC and duodenal endoscopy performed later showed microlithiasis and periampullary duodenal diverticulum with extrinsic compression of the common bile duct. Figure 1A-B showed the findings of duodenal endoscopy.

These findings allowed a diagnostic of Lemmel's syndrome; as phincterotomy was performed during the endoscopy as the treatment of choice. The patient's recovery was uneventful, and hecontinued his follow-up in the outpatient clinic surgery. After endoscopy, reassessment of the CT and MRC allowed the identification of the initially missed Lemmel's diverticulum (Fig. 2A-D).

Discussion.

Historical data.

Duodenal diverticula were first described in 1710 by the French pathologist Chomel [6]. In 1934 Lemmel was the first to report juxtapapillary diverticular and hepato-

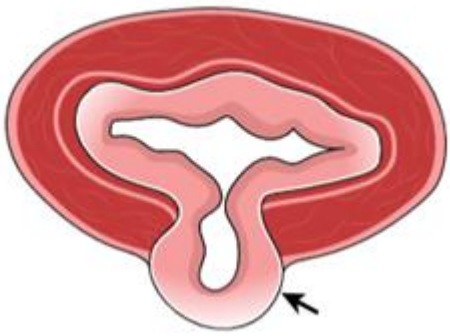


Fig. 3 a (Рис. 3 а)

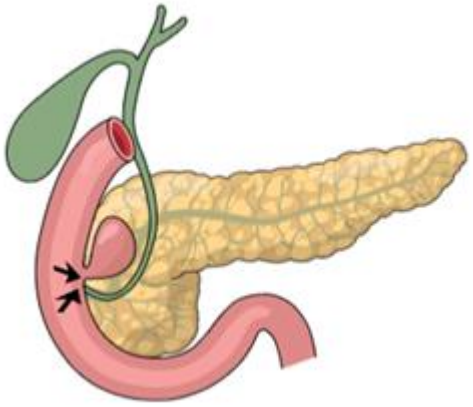
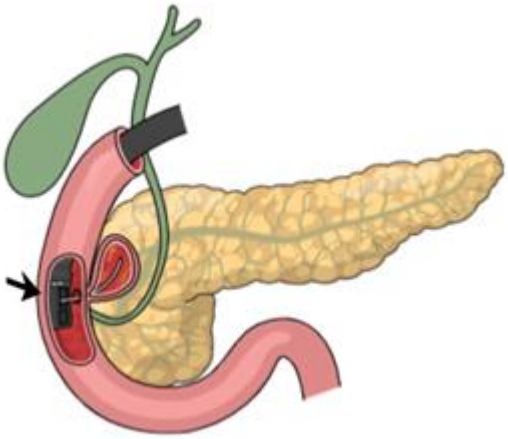


Fig. 3 b (Рис. 3 б)



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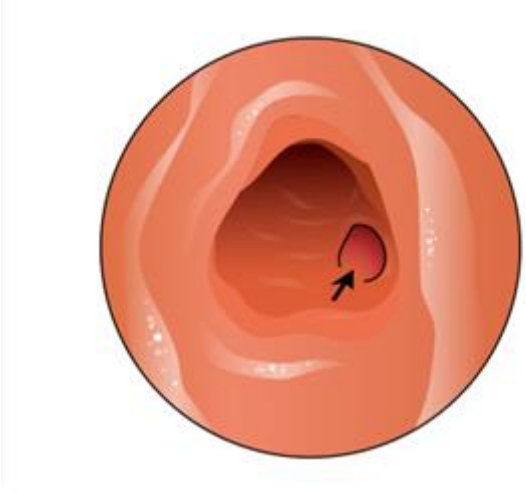

Fig. 3 d (Рис. 3 г)

Fig. 3. Diagram of the duodenal diverticulum (Lemmel's syndrome).

A – protrusions of the intestinal wall with a sac-like structure defining a gastrointestinal diverticulum (black arrow). B – a dilated duodenal diverticulum will cause common bile duct obstruction (black arrows). C, D – endoscopic view of an intraluminal diverticulum in the duodenal wall, showing the location within 2 to 3 cm to the ampulla of Vater (black arrows).

Рис. 3. Диаграмма. Дивертикул двенадцатиперстной кишки (синдром Леммеля).

А –выпячивания кишечной стенки мешковидной формы, являющиеся дивертикулами желудочно-кишечного тракта (черная стрелка). В – расширенные дивертикулы двенадцатиперстной кишки вызывают обструкцию общего желчного протока (черные стрелки). С, D – эндоскопическая картина внутрипросветного дивертикула в стенке двенадцатиперстной кишки, расположенного в пределах 2–3 см от фатеровой ампулы (черные стрелки).

cholangiopancreatic disease, excluding cholelithiasis in the absence of gallstones [7, 8].

Definition.

Gastrointestinal diverticula are protrusions of the intestinal wall with a sac-like structure that can appear anywhere in the gastrointestinal tract.

The most frequent place where diverticula are located is in the colon, followed by the duodenum [5, 9].

Lemmel's syndrome is defined as obstructive jaundice caused by periampullary duodenal diverticula (PAD) in the absence of choledocholithiasis or tumour due to biliary me-

chanical compression and can be recurrent. It is usually shown as a cavitary lesion of the medial wall of the second or third duodenal portion, either containing gas or fluid, causing obstruction of the distal part of the common bile duct and dilation of the biliary tract above. [5, 10]. Duodenal diverticula are primarily asymptomatic and found incidentally in up to 22% of the population [7].

Aetiology and classification.

The duodenum diverticula are located more frequently in the second portion of the duodenum (up to 75%), close to the ampulla of Vater (juxtapapillary) due to the wall's weaknessin this area, which arises within 2-3 cm of the ampulla of Vater [3, 6]. Cases can be classified as pancreaticobiliary or non-pancreaticobiliary [6]. It can also be classified as extramural or intramural diverticula, most of which are extra-luminal duodenal diverticulum (EDD). EDD is a herniation acquired from a defect in the bowel wall due to the entrance of vessels (Fig. 3). Besides, intramural duodenal diverticulum (IDD) is a rare congenital anomaly resulting from incomplete canalization of the lumen [7].

The intraluminal duodenal diverticulum is an uncommon congenital disability caused by inadequate intestinal lumen cannulation. Periampullary diverticula, peripapillary diverticula, and perivaterian diverticula are all terms for duodenal diverticula, located 3 cm from the ampulla of Vater. Diverticula that do not contain papilla are also named juxtapapillary or juxtampullary [11].

According to their origin, they can also be classified as congenital, which have all the layers of the duodenal wall and are generally associated with malformations or pancreatobiliary alterations, and acquired diverticula (pseudodiverticula, lacking a muscular layer), developed by a progressive weakening of the smooth muscle [6].

Several abnormal mechanisms have been proposed as possible etiologic factors in Lemmel's syndrome. The first one, diverticulitis or direct mechanical irritation of periampullary duodenal diverticulum resulting in chronic inflammation of ampulla, can lead to chronic fibrosis of papilla. Second, the periampullary duodenal diverticulum is distended by enterolith or bezoar, resulting in obstructive jaundice [4].

Pathophysiology.

Dependent on the location of the PAD, several situations can develop: chronic fibrosis of the papilla might occur secondary to periampullary diverticulitis and chronic inflammation of the ampulla. ThePAD area may cause the sphincter of Oddi to malfunction, leading to a functional obstruction. Alternatively, obstructive jaundice can result from external compression of the common bile duct or the ampulla of Vater by a PAD filled with either inflammatory debris, such as pus or enteroliths [6].

The pathophysiology can be explained with three fundamental processes, which can be successive or simultaneous:

-Direct mechanical irritation of the periampullary diverticulum can cause chronic inflammation of the ampulla, leading to fibrosis.

-Periampullary diverticula cause dysfunction of the sphincter of Oddi, resulting in stasis and bile reflux from the duodenum to the bile duct.

-The periampullary diverticulum can compress the distal part of the common bile duct or ampulla [6].

In the absence of an appropriate muscle layer or heterotrophic pancreatic tissue, the pathogenesis may develop through a site with a lack of resistance that forms through the duodenum wall at the passage of the biliary and pancreatic duct and blood vessels [12].

Clinical manifestation

Roughly 5% of the patients had any symptoms. Most Lemmel's syndrome patients develop jaundice, abdominal pain, or acute cholangitis, and these symptoms might appear intermittently [11].

Clinical symptoms consist of right upper quadrant pain, and laboratory workup would reveal elevated bilirubin levels, elevated liver enzymes, or pancreatic enzymes depending on the involvement of the ampulla of Vater. Leukocytosis, high inflammatory markers, such as erythrocyte sedimentation rate and C-reactive protein, elevated direct and total bilirubin, liver enzymes, alkaline phosphatase, and gamma-glutamyl transferase [6].

Lemmel's syndrome can cause bile stasis cholangitis. With the discovery of the Enterobacteriaceae Edwardsiella, mild to severe cases associated have been recorded and uncommon pathogen findings [13].

Diagnosis.

Duodenum diverticula are most frequently asymptomatic, and the diagnosis is confirmed by imaging, including barium meal, EUS, endoscopic retrograde cholangiopancreatography, CT, or magnetic resonance cholangiopancreatography [2].

Imaging findings.

Imaging is essential for accurate identification, and understanding imaging methods can help avoid mishandling of the duodenal diverticulum [4]. Ultrasonography, CT scan, and MRCP are available techniques to confirm

Table №1. Noda'sendoscopic classification for diverticula.

Diverticulum	Papilla-diverticulum relationship
Type A	The greater papilla is located away from the duodenal diverticulum.
Type B	The papillais located adjacent to the diverticulum.
Type C	The papillais situated on the margin of the diverticulum.
Type D	The papillais located within the diverticulum.

Table №2. Boix'sendoscopic classification for diverticula.

Diverticulum	Papilla-diverticulum relationship	
Type I	The papillais located within the diverticulum.	Ia: above. Ib: left. Ic: under. Id: right.
Type II	The papillais situated on the margin of the diverticulum.	IIa: left apical margin. IIb: right apical margin. IIc: margin centre. IId: between two diverticula.
Type III	The papillais located near the diverticulum.	

a diagnosis [10].

Ultrasonography: is frequently the first imaging method used in the emergency department to identify indirect indicators of biliary obstruction, such as biliary duct dilatation [10].

Duodenal barium study: PAD appears as contras-filled outpouchings, originating from the medial aspect of the duodenum in barium study. These diverticula can often get filled with fluid and be misdiagnosed as a pancreatic abscess, cystic tumour in the pancreatic head, or a metastatic lymph node [14].

Due to its quick acquisition and availability, a CT scan is commonly the first imaging modality used to either lead or confirm a diagnosis. CT can be performedwith intravenous contrast enhancement or orally ingesting a contrast solution [10]. It can show cystic or solid lesions on the medial wall of descending duodenum, often mistaken for pancreatic pseudocyst, abscess, or cystic neoplasm. The periampullary duodenal diverticulum also compressed the pancreatic head, but no associated pancreatic duct dilatation [4].

MRCP is the gold standard for confirming the diagnosis can distinguish diverticulum from pseudocyst or cystic tumours of the pancreatic head[10]. Side viewing endoscope during ERCP is considered the gold-standard diagnostic test, showing a primarily filled defect within the dilated periampullary duodenal diverticulum [4, 7].

There are two endoscopic classifications

Table №3. Li-Tanaka endoscopic classification for diverticula.

Type I	Papilla has an intradiverticular location, and it is not located next to the edge of the diverticulum.
Type II	The papilla is located at the edge of the diverticulum or less than 1 cm outside of the edge. IIa. Inside the edge. IIb. Less than 1 cm outside the edge.
Type III	Its location is more than 1 cm outside the edge.
Type IV	Two or more diverticula are present. IVa. The papilla is located at the edge of one of the diverticula. IVb. The papilla is located more than 1 cm from both diverticula.

for diverticula: the Noda and the Boix, tables 1 and 2.

They serve as support to plan the ERCP procedure and predict failure and difficulty of the cannulation of the papilla and complications [6]. Currently, the Li-Tanaka classification has been proposed (Table 3).

Differential diagnosis.

Diagnosing Lemmel's syndrome is often very challenging due to many other causes of obstructive jaundice [7]. The differential considerations included pancreatic pseudocyst, infected necrotic collection related to pancreatitis, paraduodenal pancreatitis, periampullary neoplasm, head of the pancreas neoplasm, metastatic lymph node, and Todani type II choledochal cyst [4].

Treatment.

Because PAD is frequently asymptomatic, treatment is only necessary when the patient becomes symptomatic. Endoscopic treatment, such as papillary balloon dilatation or endoscopic sphincterotomy with biliary stent insertion, is the treatment of choice, with a high recurrence rate [10].

The treatment of a duodenal diverticulum is determined by its clinical appearance. Asymptomatic patients are often not managed in these situations unless complications arise. In oligosymptomatic patients, conservative treatment includes nasogastric decompression and, in the event of perforation, a broad-spectrum antibiotic therapy [11].

Most patients presenting with symptoms indicative of biliary obstruction or cholangitis, endoscopic extraction, extracorporeal shockwave lithotripsy, or surgery, such as a diverticulectomy, may be indicated. If Lemmel's syndrome is due to chronic papillary fibrosis or dysfunction of the sphincter of Oddi, then an endoscopic sphincterotomy is preferred [6].

Endoscopic sphincterotomy, papillary balloon dilatation, and conservative medical management is the treatment of choice for biliary or pancreatic complications [3, 7].

Diverticulectomy is always associated with a high morbimortality due to biliary pancreatic tract lesion risk; an enterogastric derivation is preferred, especially if there is a local inflammation or diverticular perforation risk [11].

Surgery is recommended during the circumstance of endoscopic treatment failure, which may include a trans-duodenal diverticulectomy or an end-to-side Roux-en-Y choledochojejunostomy; unfortunately, these surgeries are complicated and have high mortality [10].

Complications.

Common complications include bacterial overgrowth and the production of gallstones due to beta-glucuronidase activity inducing sphincter of Oddi dysfunction by conducting a stasis and bile reflux from the duodenum to the bile duct, acute pancreatitis, or cholangitis [3, 8]. Pancreaticobiliary complications of PAD centrearound the obstructive capabilities of the diverticulum and can manifest as obstructive jaundice, cholangitis, or pancreatitis. Non-pancreaticobiliary complications include bleeding, fistula formation, perforation, or enterolith, secondary to inflammation [6].

In conclusion, Lemmel's syndrome is a rare cause of biliary obstruction. Mainetiologies include direct mechanical irritation of the periampullary diverticula, dysfunction of the sphincter of Oddi, and mechanical compression of the distal common bile duct. Diagnosing Lemmel's syndrome in patients with periampullary diverticula is extremely important in avoiding delays in diagnosing and managing these patients. In a barium study, the duodenal diverticulum appears as contrast-filled outpouchings arising from the medial aspect of the duodenum. A cystic or solid lesion on the medial wall of the descending duodenum can be misinterpreted as a pancreatic pseudocyst, an abscess, or a cystic neoplasm on CT. The diverticulum can be distinguished from pseudocysts or cystic tumours of the pancreatic head using MRC. A side-viewing endoscope during duodenal endoscopy, on the other hand, is regarded as the gold-standard diagnostic test. In cases of obstructive jaundice without choledocholithiasis or tumour, it should be evaluated as a differential. Ignoring this likelihood can result in repeated jaundice and possibly cholangitis, increasing the patient's risk of mortality and morbidity. Clinicians should be aware that 1% to 2% of periampullary duodenal diverticula may become symptomatic, causing acute abdominal pain; this clinical suspicion supplemented with imaging evaluation may prevent misdiagnosis of biliopancreatic colic due to pancreatitis or obstructive jaundice.

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this article.

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