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Lemmel Syndrome: A Case Study and Literature Review

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Abstract

The research objective was to describe the current state of knowledge about Lemmel Syndrome. Methodology. This research focused on an exhaustive literature review and a clinical case report of Lemmel Syndrome. The literature review was conducted using databases such as PubMed, EMBASE, and other relevant sources, including MeSH terms, DeCS terms, and specific keywords related to the study dimensions. The clinical case involved a 58-year-old female patient who presented to Hospital Luis Vernaza in Guayaquil, Ecuador, with an anatomical history of a juxtapapillary diverticulum. The patient presented with a 12-hour history of diffuse and intense abdominal pain, unresponsive to oral analgesics, accompanied by nausea, vomiting, fever (38°C), and general malaise, raising suspicion for Lemmel Syndrome. Results and Discussion. Lemmel Syndrome, while infrequent, should be considered as a differential diagnosis in patients with gastrointestinal symptoms. The pathophysiology of this condition, related to compression of the common bile duct by a juxtapapillary diverticulum, justifies the central role of ERCP as the diagnostic and therapeutic tool of choice. While conservative management and ERCP are often sufficient to resolve biliary obstruction, surgery may be necessary in cases of complications or lack of response to endoscopic treatment. Advances in endoscopic techniques and materials suggest a promising future for individualized and timely treatment of this and similar pathologies.

Keywords: *Lemmel Syndrome, Obstructive Jaundice, Yuxtapapillar Diverticulum.*

Introduction

Lemmel Syndrome is defined as obstructive jaundice caused by a periampullary duodenal diverticulum, in the absence of choledocholithiasis or neoplasms. It is named after the German physician Von Gerhard Lemmel of the University of Leipzig in 1934 (1), who, by studying and analyzing the case histories of different hospitals around the world from 1899 to 1933, arrived at this definition.

To date, very few cases have been published, making it a rare pathology. In Argentina, case reports have been published, such as the cases of Vega or Fernando Daza, who highlight the diagnosis of Lemmel syndrome through Magnetic Resonance Imaging studies (2,3).

Diverticula located in the small intestine correspond to a finding of 2 to 5.76% in imaging studies, reaching 20% as findings in autopsies; and are most frequently located in the duodenum with 79%, explicitly in the second portion of the duodenum, adjacent to the ampulla of Vater or at a distance of 2-3 cm (2.3). Most are extraluminal and 90% are solitary (4).

Although duodenal diverticulum is a common incidental finding in the general population, it is rarely symptomatic. When this happens (5% of cases), it is usually overlooked or presents a diagnostic and therapeutic challenge. With the scope of endoscopic studies and the increase in the age of the population, a variable prevalence has been recorded ranging between 5-10%, and a higher prevalence in older people, reaching 27% in octogenarians. No differences have been found regarding sex.

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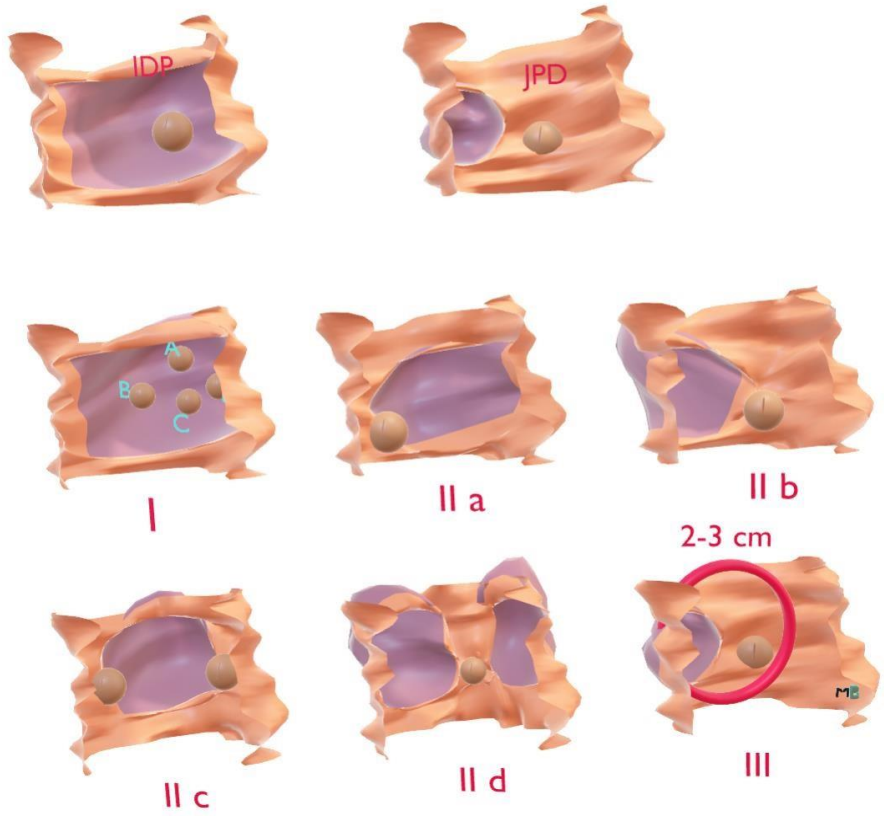
When symptoms occur, they are usually very nonspecific, such as pain or abdominal discomfort, up to malabsorption that may be caused by bacterial overgrowth (5,6). The presence of significant symptomatology may be caused by the size of the diverticulum, its location, and its relationship with the structures that are near it; these symptoms may be: intestinal obstruction, infection, jaundice, abdominal pain, perforation or bleeding.

Diverticula that occur in the small intestine usually remain asymptomatic and their detection is incidental (5–7). According to Hasenfuß (2021), who mentions that digestive hemorrhage caused by diverticula in the small intestine is usually produced by those located in the third and fourth portion of the duodenum, which is related to the presence of gastric acid and ectopic gastric mucosa.

The appearance of jaundice, cholangitis or acute pancreatitis develops in the presence of ampullary, juxtapapillary and peripapillary duodenal diverticula, leading to obstruction or infection in this area, the papilla of Vater and the distal bile duct (7). Diverticula located in the jejunum and ileum are generally asymptomatic and are identified incidentally by imaging techniques, surgeries or autopsies (8).

The pathophysiology in small bowel diverticula is not yet very well detailed, although they may be caused by muscle dysfunction and the myenteric plexus; some authors report that alterations in peristalsis, intestinal dyskinesia, which leads to increased pressure in localized areas of the small intestine, may be involved, therefore the increase in intraluminal pressure may be one of the reasons, and even inflammation that may be associated with bacterial overgrowth in the small intestine (5–7,9).

Figure 1. Boix and Lobo Classification



Fuente: Yue et al (10).

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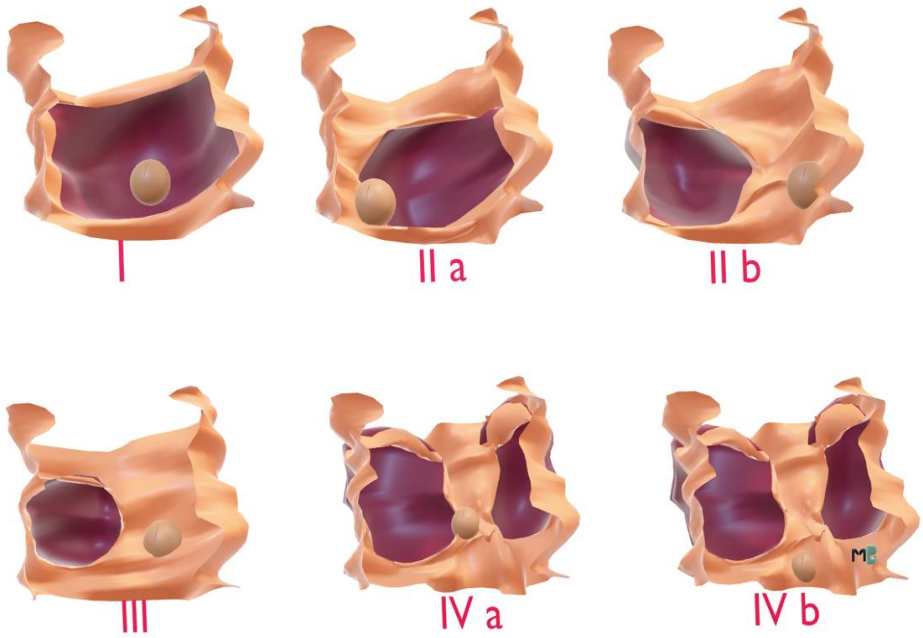
Figure 2. Li-Tanaka Classification



Fuente: Yue et al (10).

Three classifications have been made based on the arrangement of periampullary diverticula; the classification of Lobo Yue et al. is one of them (10).

- Intradiverticular papilla: The major papilla is located within the diverticulum.
- Juxtapapillary diverticulum: The papilla is located outside the diverticulum.

Boix Classification (10) (see Figure 2)

- Type I: The papilla is located within the diverticulum.
  - ✓ Ia: Superior
  - ✓ Ib: Left
  - ✓ Ic: Inferior
  - ✓ Id: Right
- Type II: The papilla is located at the margin of the diverticulum.
  - ✓ IIa: Left apical margin
  - ✓ IIb: Right apical margin
  - ✓ IIc: Center of the left or right margin
  - ✓ IId: Between two diverticula

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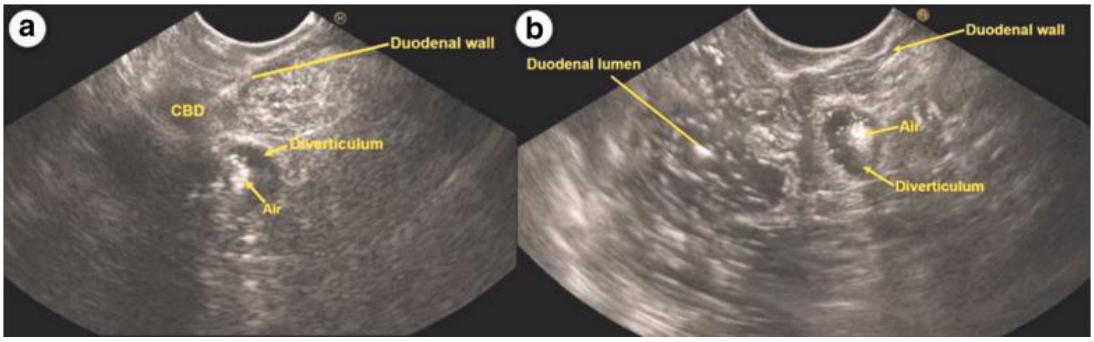
- Type III: The papilla is located near the diverticulum.
  - ✓ Li-Tanaka Classification (10) (see Figure 3)
  - ✓ Type I: The papilla is located within the diverticulum and not adjacent to the margin.
  - ✓ Type II: The papilla is located at the margin of the diverticulum:
  - ✓ IIa: Within the margin
  - ✓ IIb: Outside the margin, < 1 cm
- Type III: The papilla is located outside the margin, ≥ 1 cm
- Type IV: The papilla is located at the margin of the diverticulum, ≥ 2 diverticula
  - ✓ IVa: The papilla is located outside the margins of at least one diverticulum, < 1 cm
  - ✓ IVb: The papilla is located outside the margins of all diverticula, ≥ 1 cm

According to Jakubczyk (2020) and Ping Yue (2020), the most frequent intradiverticular location was type II according to the Li-Tanaka classification (10,11) (Figure 2).

Diagnosis is usually an incidental finding on an upper gastrointestinal contrast study showing the "duodenal windsock sign" (contrast-filled sac surrounded by a thin line of radiotranslucency), whereas the diagnosis of an extraluminal diverticulum is usually made by contrast retention for more than 6 hours in oblique views in the upright and recumbent positions, with the presence of a narrow neck that differentiates it from a bulbar or postbulbar duodenal ulcer in D1 and D2 respectively (12).

The use of linear echoendoscopy has been reported as a diagnostic method for the evaluation of the dilated common bile duct. During the procedure, an abnormal soft tissue is observed extending towards the dilated distal common bile duct, which contains multiple echogenic air images suggestive of a diverticulum (Figure 4). After instillation of water, the diverticulum increased in size, water together with air bubbles (echogenic foci) that move from the duodenal lumen to the diverticulum (Figure 5) and resulted in obvious compression of the biliary tract that led to the diagnosis of Lemmel syndrome (13).

Figure 3. A) Linear Echoendoscopy Showing a Periampullary Diverticulum Containing Air That Compresses the Lower Common Bile Duct. B) Echoendoscopy In the Second Portion of the Duodenum Showing A Duodenal Diverticulum Containing Echogenic Air Image Near The Ampulla.



Fuente: Somani & Sharma (13).

The gold standard technique for diagnosis is Endoscopic Retrograde Cholangiopancreatography (ERCP), which has proven to be a useful method for both diagnosis and treatment of pancreatobiliary conditions.

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With the advances that have been made, new endoscopic techniques and materials, ERCP is today a safe and effective procedure, which promises to resolve even complications such as perforations. However, several complications continue to be documented (13,14).

Figure 4. Position of Duodenal Diverticulum Within A Range of 2 To 3 Cm Distance from the Major Duodenal Papilla

Magnetic resonance cholangiopancreatography (MRCP) is a valid imaging method for evaluating the biliary tree, as well as detecting coexisting abnormalities. It has proven useful as a method of direct visualization of the effects of a duodenal diverticulum on the common bile duct, but not for the assessment of the diverticulum per se or its complications. One advantage is that it is a non-invasive method. In addition, it provides a superior delineation of the relationship between the diverticulum and the papilla, allows us to better characterize the anatomy of the common bile duct, pancreatic duct, and pancreatic parenchyma (12). Therefore, in this type of diagnostic technique; small diverticula may be easily missed, as may those that do not contain fluid, so the joint administration of water and changes in the patient's position within the examination process would be recommended. This fluid could also be increased after stimulation with secretin from the exocrine part of the pancreas. Deviation of the common bile duct is the main criterion to deepen an investigation of the descending portion of the duodenum in search of a diverticulum, so, if this were to end directly on a diverticulum or cross it, it would be difficult to establish a specific association (15).

Surgical intervention becomes indispensable, which includes diverticulectomy and two-layer closure, by hand suture or with staples. If the duodenum is friable and there is intense surrounding peridiverticulitis, it is mentioned to add a duodenal diverticulization procedure. The operative mortality is 20% to 30%, while morbidity is 30% to 40%. In addition, a late diagnosis of perforated diverticulum or late surgical intervention, when treatment by surgery was indicated, has a mortality rate greater than 90% (12–14). Only 1% to 5% of cases of duodenal diverticula will require surgical intervention (12,16). In asymptomatic or mildly symptomatic patients, elective surgery carries a high morbidity and mortality, diverticulectomy presents with a 50% clinical success rate, so it is not considered an option; however, in the presence of serious complications such as biliary obstruction, hemorrhage or perforation or in the event of failure of conservative treatment, surgical intervention should be considered (12,17).

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ERCP is the best treatment option in these cases. It has proven to be a safe and minimally invasive technique. However, it is not without complications such as: perforation or bleeding. The presence of duodenal perforation (0.8-1%) could be favored by the difficulty generated by juxtapapillary diverticula at the time of cannulation.

ERCP allows for sphincterotomy and placement of a biliary stent; with resolution of the pathology, but these patients require observation and follow-up and evaluate when surgical approach is required (16).

In addition, ERCP is useful by offering intraluminal diverticulectomy, the location and control of hemorrhagic duodenal diverticula can be performed; however, a high number of failures, and a high rate of rebleeding, as well as high rates of mortality and morbidity have been reported. Angiography would allow to locate and control the episode of hemorrhage not controlled by endoscopy.

If there is failure in the management of symptoms or endoscopic treatment, surgical exploration and diverticulectomy should be considered as an option (5), with the advances described so far in the management of complications and new endoscopic techniques and supplies, make the procedures possible and safer.

Iatrogenic duodenal perforations by ERCP are possible complications that could occur during treatment, these are very rare approximately 0.8-1% of cases and more frequent when they are near the papilla, as is the case of this pathology (18). We must bear in mind that their recognition and treatment is of great importance because it will affect the patient's prognosis, for this the type of perforation will be identified by Stapfer scale and the most indicated treatment (19).

Another advance in endoscopic treatment is the use of mechanical sutures, which have their use primarily in bariatric surgery, could be taken as a solution in complicated diverticular processes in which endoscopic treatment could be performed. Endoscopic suturing has some applications such as stent fixation and closure of larger defects, such as fistulas and perforations, but it is technically more demanding. OverStitch (Apollo Endosurgery Inc., Austin, TX, USA) is a single-use disposable suture device that uses a double-channel therapeutic endoscope to apply continuous or intermittent stitches without removing the endoscope to reload the needles. Tissue approximation is facilitated by a tissue retraction device or grasping forceps and full-thickness suture is facilitated by a helical tissue catheter. Sharaiha et al in their study showed that endoscopic suturing used for stent fixation and closure of fistulas and perforations achieved high technical (97%) and clinical 1 (79%) success. Clinical success was maintained for 1 to 3 months of follow-up without the need for additional intervention. While primary closure of acute fistulas (less than 30 days) after bariatric surgery has been very successful, closure of chronic fistulas is disappointing with a success rate as low as 23%; therefore it could be considered as a therapeutic option in the presence of duodenal diverticula (19,20).

Methods

This research focused on an exhaustive literature review and a clinical case report of Lemmel Syndrome, with the objective of describing the current state of knowledge about this condition. The literature review was conducted using databases such as PubMed, EMBASE, and other relevant sources, including MeSH terms, DeCS terms, and specific keywords related to the study dimensions such as: "Lemmel Syndrome", "Lemmel Syndrome", "Obstructive jaundice", "Obstructive jaundice", "Juxtapapillary diverticulum", "Juxtapapillary diverticulum". Articles published in recent years that address Lemmel Syndrome will be selected, including case studies, case series, reviews and meta-analyses.

The case of a 58-year-old female patient with a history of hypertension, laparoscopic cholecystectomy and hysterectomy, and an anatomical history of juxtapapillary diverticulum, who was treated at the Luis Vernaza Hospital in Guayaquil, Ecuador, was presented. The patient's clinical history was detailed, including symptoms, laboratory findings and imaging studies, as well as the diagnostic procedures, which led to the diagnosis of Lemmel Syndrome, as well as the therapeutic treatment performed.

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Clinical Case

A 58-year-old Ecuadorian female patient, treated at the Luis Vernaza Hospital in Guayaquil, Ecuador, with a medical history of hypertension treated with losartan and hydrochlorothiazide, laparoscopic cholecystectomy, hysterectomy, and an anatomical history of juxtapapillary diverticulum, presented with a 12-hour history of diffuse and intense abdominal pain, unresponsive to oral analgesics. It is accompanied by nausea, vomiting, fever (38°C), and general malaise. Laboratory findings were as follows:

Table 1. Admission Laboratory Tests

		Reference Range
White Blood Cell Count	11.09 x 10^3/uL	4.40 – 10 x 10^3/uL
Neutrophils	54%	50 – 70%
Hemoglobin	12 g/dL	12.6 – 16.4 g/dL
Hematocrit	36.1%	38 – 48%
Platelets	329 x 10^3/uL	150 - 450 x 10^3/uL

		Reference Range
Total Bilirubin	1.69 mg/dL	0 - 1 mg/dL
Direct Bilirubin	1.30 mg/dL	0 - 0.3 mg/dL
Indirect Bilirubin	0.39 mg/dL	0 – 0.7 mg/dL
AST (GOT)	256 U/L	0 – 32 U/L
ALT (GPT)	740 U/L	0 – 31 U/L
GGT	671 U/L	7 – 32 U/L
Alkaline Phosphatase	463 U/L	35 – 104 U/L
Amylase	230 U/L	28 – 100 U/L
Creatinine	1.06 mg/dL	0.5 – 1.3 mg/dL
Urea	40 mg/dL	6 – 24 mg/dL
Glucose	110 mg/dL	70 – 100 mg/dL

Abdominal ultrasound shows dilation of the common bile duct (1.3 cm), slightly dilated intrahepatic biliary tree, which is inconclusive, so a magnetic resonance cholangioresonography is requested, which is of normal characteristics.

The condition is interpreted as "cholangitis without a clear obstructive cause", antibiotic treatment with ciprofloxacin and metronidazole is decided.

Due to the anatomical history of juxtapapillary diverticulum with current presence of cholestasis laboratory; ERCP is scheduled with the diagnostic suspicion of Lemmel syndrome.

During this procedure, cannulation of the difficult papilla was observed, due to severe fibrosis and the location of the duodenal diverticulum. However, cannulation of the papilla with sphincterotomy is achieved (Fig 5).

The patient progresses with clinical improvement and decreasing liver enzymes.

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Figure 5. Endoscopic Retrograde Cholangiopancreatography (ERCP). After Successful Cannulation, Sphincterotomy of the Ampulla of Vater is Observed.

Five years later, she is again admitted to the emergency area presenting a clinical picture characterized by abdominal pain of moderate intensity in the epigastrium with irradiation to the mesogastrium, vomiting of food content and generalized jaundice and a new pattern of cholestasis by laboratory.

A cholangioresonance is performed, where dilation of the intra and extrahepatic biliary tract and an isointense image in the distal common bile duct, which speaks of partial distal stenosis, are observed.

Using the lateral vision duodenoscope, it is advanced to the second portion, where a large diverticulum is observed in the region of the papilla, which has abundant retained food content.

With the objective of draining the biliary tract, the placement of a 2 cm self-expanding prosthesis + 5 cm plastic stent is decided through ERCP, achieving adequate biliary drainage. It evolves with clinical and analytical improvement.

One month later, she comes to check-up where migration of the plastic stent is observed. It is removed, leaving only the metal prosthesis, which was removed in the following 2 weeks. (Figure 6). The patient is discharged from the hospital and continues to be assessed on an outpatient basis.

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Figure 6. Lateral View of the Duodenoscope. the Duodenal Diverticulum Protruding the Ampulla of Vater (White Arrow) is Observed, As Well As the First Duodenal Portion (Green Arrow), and the Second Duodenal Portion (Blue Arrow).

Months later, she again presents with a clinical picture: moderate to severe colicky abdominal pain located in the epigastrium that radiates to the right hypochondrium and right iliac fossa that does not subside with analgesics, accompanied by nausea, vomiting of bilious content, febrile (39 ° C), hypotension 90/60 mmHg, MAP 70 mmHg, tachycardia 120 bpm. Laboratory:

Table 2. Laboratory Findings Third Admission

		Reference Range
White Blood Cell Count	15.19 x 10^3/uL	4.40 – 10 x 10^3/uL
Neutrophils	91.9%	50 – 70%
Hemoglobin	10.6 g/dL	12.6 – 16.4 g/dL
Hematocrit	31.0%	38 – 48%
Platelets	255 x 10^3/uL	150 - 450 x 10^3/uL

		Reference Range
Total Bilirubin	1.48 mg/dL	0 - 1 mg/dL
Direct Bilirubin	1.18 mg/dL	0 - 0.3 mg/dL
Indirect Bilirubin	0.31 mg/dL	0 – 0.7 mg/dL
AST (GOT)	98 U/L	0 – 32 U/L
ALT (GPT)	201 U/L	0 – 31 U/L
GGT	150 U/L	7 – 32 U/L
Alkaline Phosphatase	100 U/L	35 – 104 U/L
Amylase	1464 U/L	28 – 100 U/L

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Creatinine	0.59 mg/dL	0.5 – 1.3 mg/dL
Urea (BUN)	43.6 mg/dL	6 – 24 mg/dL
Glucose	91 mg/dL	70 – 100 mg/dL
Sodium (Na+)	138 mEq/L	133 – 145 mEq/L
Potassium (K+)	4.5 mEq/L	3.5 – 5.4 mEq/L

She is hospitalized with a presumptive diagnosis of "acute alithiasic pancreatitis" BISAP 2 points, SOFA 3 points, Apache II 7%.

At this point we find ourselves in the context of a patient with a history of Lemmel syndrome, which had an initial clinical improvement with conservative treatment, and which evolves with multiple occurrences: cholangitis, pancreatitis.

A valuation by the surgery service is decided, who decide to perform an exploratory laparotomy, because it is a patient with the presence of a parabacterial duodenal diverticulum (with food retention) that causes symptoms of obstruction of the extrahepatic bile duct repeatedly and did not resolve with endoscopic treatment.

Surgery is performed without complications in which a free vesicular bed is visualized, the main bile duct dilated in its entire length. No externalized periampullary diverticulum is observed, but rather directed towards the pyloric region through the internal face, it is decided to make a anastomosis in Y of roux, to then establish the hepatoyeyunal biliodigestive anastomosis.

After the surgical intervention, a gastroduodenal transit was performed; where the persistence of the duodenal diverticulum is evidenced (Figure 7-8).

Figure 7. Gastrointestinal Transit. The Barium Passage Highlights a Diverticular Structure at the Duodenal Level (Green Arrow).

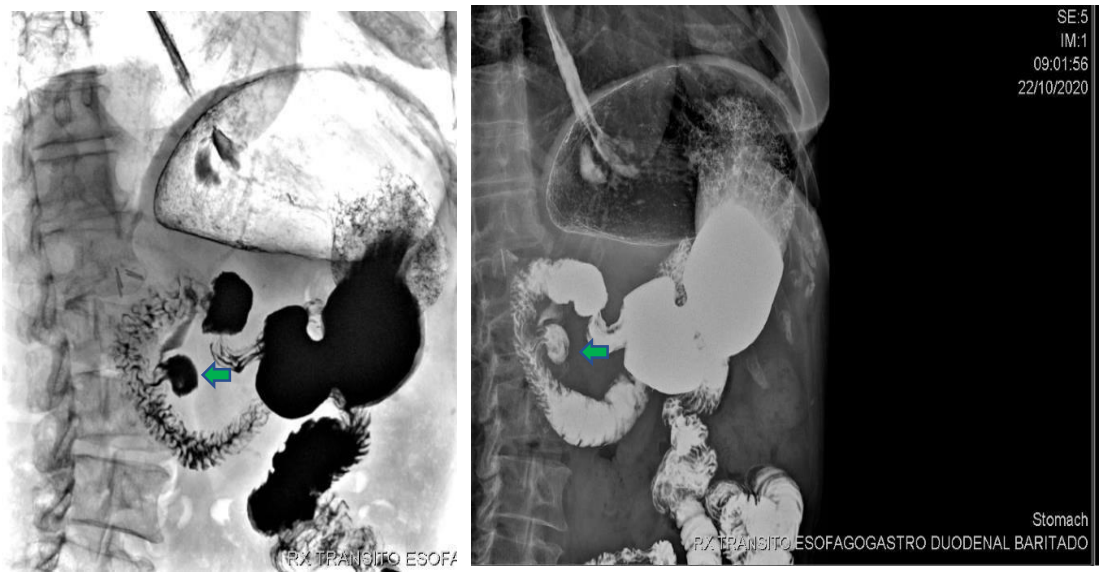

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Figure 8. Gastrointestinal Transit. Follow-Up After One Year,

Reflecting Measurements of The Duodenal Diverticulum Of 18.4 Mm (Blue Arrow).

This is a very well-written and comprehensive discussion of Lemmel Syndrome! Here's a translation into English that aims to preserve the clarity and detail of your writing:

Discussion

As specified in the bibliographic review carried out; the finding of diverticula in the small intestine is infrequent and even detected incidentally, with the most frequent site of location being the second portion of the duodenum.

The presence of obstructive jaundice caused by a periampullary duodenal diverticulum and in the absence of choledocholithiasis or neoplasms is called Lemmel syndrome; the diverticulum causes direct external compression of the common bile duct which leads to obstruction; causing irritation of the ampulla with its subsequent chronic inflammation and fibrosis of the papilla; as evidenced in the clinical case.

The symptomatology is infrequent and in case of presenting clinical manifestations these are usually nonspecific; so its diagnosis is usually difficult and imprecise, as demonstrated in the case presented; in which the patient presented abdominal pain, nausea and vomiting, which led to her hospital admission in the emergency room on several occasions.

Azzam et al (21) mentions in his article; that the patients in his study suffered from recurrent acute abdominal pain, which required visits to the emergency services. It also showed that patients improved with the administration of antibiotics, the placement of a plastic biliary stent by ERCP and the drainage of the obstruction, respectively; as shown in our case.

In relation to the complications caused by duodenal diverticula; Aourarh et al (22); mentions that non-pancreatobiliary complications such as hemorrhage, diverticulitis, perforation or fistula formation can occur in 5% of cases; and pancreatobiliary complications such as acute pancreatitis, cholangitis, gallstones in the bile ducts or obstructive jaundice that cause Lemmel Syndrome (11); in the case described, the presence of non-pancreatobiliary complications such as diverticulitis, bacterial overgrowth, pancreatobiliary complications such as the presence of acute pancreatitis, cholangitis could be observed.

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The literature states that no treatment is required in asymptomatic patients; but constant monitoring should be carried out to avoid complications.

In our patient, a conservative approach was taken in her initial stage, going through antibiotic coverage treatments to invasive procedures: such as ERCP with sphincterotomy, prosthesis placement and surgery.

This finally managed to treat the obstruction and allowed her to remain in follow-up and without presenting complications.

Sosa indicates that the objective of treatment is to relieve biliopancreatic obstruction by surgical resection of the diverticulum, endoscopic sphincterotomy and/or endoprosthesis; but that endoscopic treatment has the highest success rate, while surgical management is reserved only for serious complications such as perforation, hemorrhage or in conditions that present with severe sepsis, and ERCP with sphincterotomy is preferable in cases of pancreatobiliary alterations such as in the presence of acute pancreatitis as presented by our patient (23). In the case we have presented, the objective of relieving the obstruction was reached, but exhausting all the resources described in the literature; with the exception that the objective of diverticulotomy is not achieved due to its difficult anatomical presentation and the risks it entails. However, the good clinical evolution of the patient is achieved.

This discussion effectively summarizes the key points of your research and highlights the importance of considering Lemmel Syndrome in patients presenting with gastrointestinal symptoms. It also emphasizes the challenges in diagnosis and the various treatment options available. The comparison with other studies and the discussion of the patient's specific case further strengthen the paper.

Conclusions

In the present work, the importance in the recognition and treatment of Lemmel Syndrome in hospital practice has been exposed, although being a pathology with low incidence, it should be considered as a differential diagnosis among usual gastrointestinal scenarios.

The bibliographic review considers that the pathophysiology in small bowel diverticula is not very well detailed, but considers several possible causes of its genesis such as muscle dysfunction and the myenteric plexus, alterations of peristalsis, intestinal dyskinesia, which leads to raise the pressure in localized areas in the small intestine, The proximity of the diverticulum to the ampulla that would cause a direct external compression of the common bile duct and lead to obstruction and can cause dysfunction of the sphincter of Oddi, among the possible causes of Lemmel Syndrome. Therefore, it would be valid to think that by compression mechanism and increased intradiverticular pressure result in the proximity of the juxtapapillary diverticulum to the distal part of the biliary tract being obstructed and consequently dilated, having a relationship with the decompression treatment that would be the ERCP as gold standard in the syndrome.

The publications consulted agree that, in the presence of asymptomatic duodenal diverticula diagnosed incidentally, no specific treatment is required, however, it is important to observe the evolution of these diverticula.

Patients who do not present limitations in their lifestyle require conservative management, without the need for surgery, with ERCP being the gold standard as a diagnostic and therapeutic option, since it allows the resolution of pancreatobiliary obstruction safely and effectively; the management of symptoms and directed therapy is one of the points of greatest interest since, as in the case presented, the management of complications is often difficult to achieve after initial treatment; but through advances in endoscopic techniques and materials that are increasingly high standards and quality, suggest that in the near future we can offer individualized and timely treatment for this or other similar pathologies.

In view of the above, it can be concluded that endoscopic management should be chosen as initial treatment, whenever it is available and probably with new techniques and materials results in greater success

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and lower mortality rate than conventional surgery. Surgery would be indicated as in the case previously exposed, if there were complications and/or lack of resolution by endoscopic method.

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