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Lemmel Syndrome as A Cause of Obstructive Biliary Syndrome Complicated with AcutePancreatitis: *A Case Report in A Hospital in Bogotá, Colombia*

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Abstract

Lemmel syndrome is defined as a form of jaundice syndrome in the absence of choledocholithiasis or neoplasms that cause bile duct obstruction, usually secondary to pseudodiverticula or diverticula that compromise the duodenal mucosa. However, to date there is no standardized consensus on the medical-surgical approach to this pathology, particularly when it comes to an incidental finding, which increases the risk of recurrence of the acute inflammatory process and mortality during hospitalization. Considering the above, the case of an 88-year-old female patient who attends the emergency service of a fourth level center in the city of Bogotá DC (Colombia), with a clinical picture compatible with acute pancreatitis secondary to Lemmel syndrome is presented. of etiological studies that ruled out more frequent pathologies. Below is a review of the literature, clinical and paraclinical findings compatible with this entity, discussion of medical management proposals according to the cases reported in the literature.

Keywords: *Lemmel Syndrome, acute pancreatitis, non-obstructive pancreatitis, duodenal diverticula.*

Introduction

Duodenal diverticula are protrusions of the digestive mucosa and submucosa through the muscular wall of the duodenum, which are caused by duodenal hyper pressure that occurs opposite the embryological line of fusion between the ventral and dorsal parts of the pancreas [1,2] Most duodenal diverticula are acquired and extraluminal, and another type described corresponds to juxtapapillary diverticula found in the second portion of the duodenum, approximately 20-30 mm from the ampulla. Finally, when the diverticulum contains the papilla, it is called an intrapulpal diverticulum. Although they can be asymptomatic in 95% of cases, in autopsy series duodenal diverticula can be found in up to 20% to 22%of the general population, and the incidence increases with age, of which less than 10 % are symptomatic [3,4].

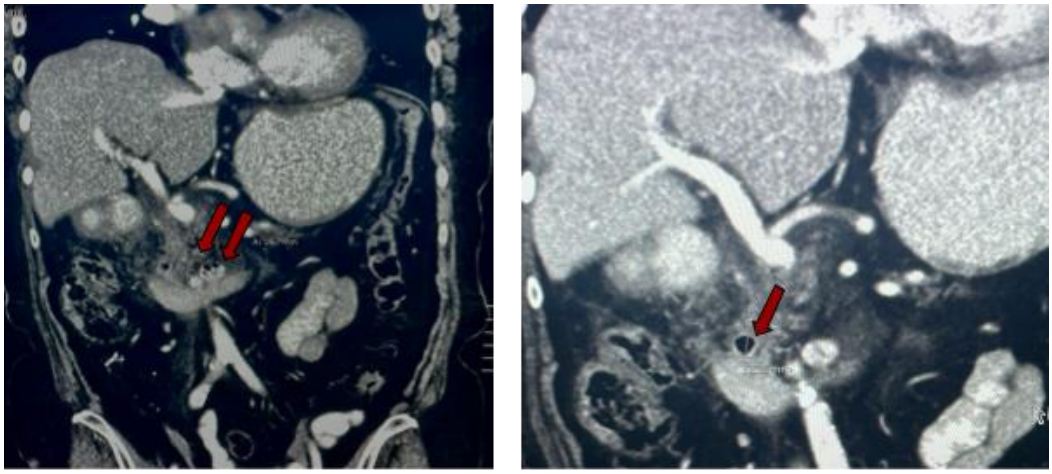
The extraluminal duodenal diverticulum (EDD) is a hernia acquired by a defect in the intestinal wall due to the entry of perforating vessels [2]. They are associated with biliopancreatic diseases such as choledocholithiasis, cholangitis and pancreatitis [1], intermittent combined pancreatobiliary obstruction can cause multiple episodes of obstructive jaundice and biliary colic with or without ascending cholangitis, as well as concomitant interval episodes of acute pancreatitis, leading to pancreatic atrophy with recurrence [6]. These diverticula have repercussions on the bile duct including mechanical obstruction, sphincter of Oddi dysfunction and a greater number of beta-glucuronidase-producing bacteria [1]. Non-biliary complications include diverticulitis, diverticular bleeding, abdominal pain, and food impaction [1, 3]. The timely diagnosis and management of this entity is essential, in order to prevent acute and chronic complications that can compromise the life of the patient in the short and medium term.

Objective:

Given the increased risk of recurrence of episodes of acute pancreatitis with multiorgan dysfunction and increased mortality and morbidity, associated with secondary complications that can occur such as perforation, chronic pancreatitis, gastrointestinal bleeding, infections among others in patients with untreated Lemmel syndrome, An unusual case in medical practice is presented, with the intention of mentioning the clinical presentation, the etiological diagnostic approach and its differential diagnoses, the proposal of clinical management. Tools that will contribute to the diagnostic approach for the medical groups that faces similar clinical cases.

Presentation of the case

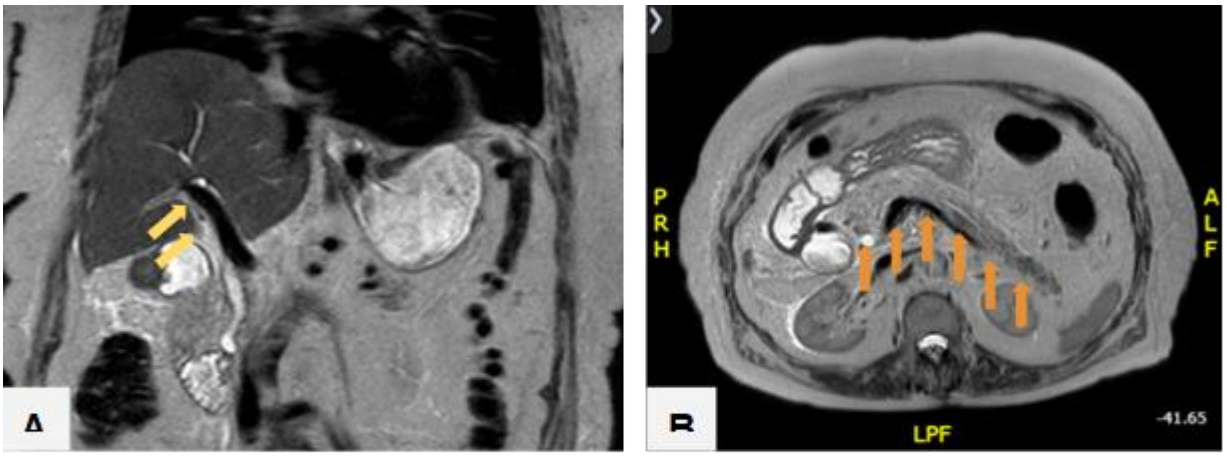
An 88-year-old female patient with a pathological history of arterial hypertension and dyslipidemia in management, and a surgical history of open cholecystectomy, who came to the emergency room due to a clinical picture of 2 days of evolution consisting of colicky abdominal pain predominantly in the epigastrium and hypochondrium. Right, of moderate intensity, radiated to the back, associated with nausea and intermittent episodes of liquid, non-dysenteric stools. However, due to exacerbation in the last48 hours, he decided to consult the physical examination of the distended abdomen, with pain onpalpation of the epigastrium, mesogastrium, and no masses or visceromegaly, without signs of peritoneal irritation. Admission paraclinics with leukocytosis of 11,600 *(N = 5,000 - 10,000)*, neutrophilia of 9,750 *(N = 2,000 - 7,000)*, without alteration of other cell lines, additional alkaline phosphatase of 273 *(N = 40- 150 U / I)*, ALT 257 *(N = 0 - 55 U / I)* and AST 731 *(N = 5 - 34 U / I)*, direct bilirubin 2.1, indirect bilirubin 1.2, total bilirubin 3.0 *(N = 0.2-1.5 mg / dL)* , amylase 564 *(N = 28 - 100 U / I)* , with elevated abnormal kidney function compatible with acute kidney injury KDIGO 2 with creatinine of 2.1 *(N = 0.6 -1.1 mg / dL)* BUN 38.91 *(N = 8 -23 mg / dL),* associated with arterial gases with uncompensated metabolic acidemia with increased GAP anion and moderate hyperlactatemia.



Appendix 1: ***Contrast computed tomography of the abdomen***, showing diverticula in the duodenum; one in the second portion of the duodenum (12.2 mm diameter), and other two in the third portion of the duodenum (8.9 mm and 14.7 mm of diameter) (red arrows)

Given these findings, it was decided to take a contrasted abdominal tomography with a report of extensive inflammatory changes in the fat adjacent to the head and uncinate process of the pancreas, and duodenum, with extension to the right flank, observing at least three duodenal diverticula; one in the second portion of the duodenum (12.2 mm diameter), and other two in the third portion of the duodenum (8.9 mm and 14.7 mm of diameter) **(see annex 1)** without clear signs of perforation. With these paraclinical and clinical findings, a classified acute pancreatitis was considered: APACHE 9 points MARSHALL 2 ICU transfer was decided, and management began with removal of the oral route, initiation of hydro saline resuscitation, analgesia, and a study was complemented with cholangioresonance in the event of alterationfindings in liver function with a report indicating dilation of the intrahepatic bile duct, without images of choledocholithiasis, confirmed extensive involvement by pancreatitis.

As an additional study, an upper gastrointestinal endoscopy was requested with evidence of severe chronic peptic esophagitis, for which a proton pump inhibitor was managed. Subsequently, an etiological study of pancreatitis was started: an obstructive origin was ruled out by means of nuclear magnetic cholangioresonance **(Annex 2)**, a patient with no history of alcohol consumption as a possible cause, a normal range for triglycerides (121 mg / dL), normal calcium (7.8 mg / dL), normal carcinoembryonic antigen (1.48 µg / L), normal alpha-fetoprotein (1.63 ng / ml). Finally, endoscopic ultrasound is performed to complement the etiological approach, ruling out neoplasia and microlithiasis. It is considered the most probable picture associated with duodenal diverticula, with a favorable clinical evolution after medical management, with recovery from multi-organ failure and compensation forunderlying pathologies, for which discharge is given.



Appendix 2: ***Cholangiomagnetic resonance imaging of the abdomen*** showing evidence of dilation of the intrahepatic bile duct **(Figure A),** and significant fat replacement of the pancreas with alteration of the signal intensity of peripancreatic fat, hepatorenal, and perirenal space **(Figure B)**

Discussion

The duodenum is the most common site of diverticula after the large intestine as found in the case of our patient, with a reported incidence of 15% to 22%. They are also described as false or true, according to the compromise of the mucosa and submucosa layers versus all the histological layers of the intestinal wall, respectively [3,4]. The pathophysiological mechanisms may involve recurrent diverticulitis or mechanical irritation of the diverticula causing inflammation around the bulla and posterior papillary fibrosis, anatomical distortion of the bulla with subsequent dysfunction of the sphincter of Oddi, and direct mechanical compression of the common bile duct or the bulla per se. by the diverticulum, which lead to a process of bile duct obstruction.

Despite its high incidence, duodenal diverticula are rarely symptomatic and are discovered incidentally on imaging or autopsies; however, complications can occur in up to 20% of patients, including bleeding, perforation and intestinal obstruction, usually associated with inflamed and / or large duodenal diverticula [1,7] These being the most frequent complications and with have the highest risk of associated morbidityand mortality. On the other hand, pancreatobiliary complications include stones impacted in the gallbladder and bile ducts, obstructive jaundice, cholangitis, as well as acute and chronic infections, including pancreatitis, a complication presented in our case. These complications are due to phenomena such as extrinsic compression of the sphincter of Oddi by the diverticulum, or the regurgitation of intestinal contents in the bile ducts, which results in an excessive growth of bacteria [2], added to the presence of bacteria in the diverticula, which favors the hyperproduction of beta-glucuronidase, an enzyme responsible for the digestion of complete carbohydrates, which has been reported as a potential cause of idiopathic acute pancreatitis [6].

Being an entity with a frequency of 5-10% of patients with diverticular disease, Lemmel syndrome should be part of the spectrum of differential diagnoses, since it can be confused with other entities such as a tumor of the head of the pancreas, with a completely different prognosis and treatment. One of the mainstays for diagnosis is through endoscopic ultrasound, which is also emerging as a therapeutic measure through endoscopic sphincterotomy with a success rate of 95% for the prevention of recurrence [5,7]. However, since it is a test of limited availability, the use of diagnostic images such as simple abdominal tomography or nuclear magnetic resonance is proposed for the identification of diverticula. In cases in which patients with duodenal diverticulum experience abdominal pain, taking amylase or lipase should be considered for the diagnosis of obstructive pancreatitis. Once diagnosed, treatment is conservative based on oral suspension in case of intolerance, fluid resuscitation and initiation of antibiotic coverage in patients with clinical or paraclinical signs of systemic inflammatory response, with adequate initial response [1].

However, it is estimated that approximately 2% of patients with Lemmel Syndrome do not respond to initial conservative management, which increases not only the risk of in-hospital mortality, but also the risk of immediate recurrence [3]. For this reason, surgical approaches have recently been proposed via exploratory laparotomy to perform diverticulectomy with subsequent sphincterotomy and ampulectomy with sphincteroplasty points (Nasser AN) [6,7]. However, the mortality of the procedure is 20-30%, and the morbidity of 30-40% [7]. Therefore, the decision of a surgical approach should be based on the presence of complications of diverticular disease (perforation, hemorrhage, etc.), which can lead to a mortality of up to 90% due to the development of generalized peritonitis.

Conclusion

The case of an 88-year-old female patient with a diagnosis of acute pancreatitis requiring hospitalization in an intensive care unit is presented, finding as a cause the presence of giant duodenal diverticula that led to obstructive biliary syndrome, an infrequent cause as previously described. For the Lemmel Syndrome approach, it is important to establish the presence of complications of diverticular disease in order to define a conservative management with the

administration of crystalloid fluids and support measures, versus the possibility of surgical management with direct manipulation of the bile duct. which can be minimally invasive by means of endoscopic ultrasound, or an open approach for the resection of diverticula. Due to its high mortality in the event of complications, it is important to consider lemmel syndrome within the spectrum of differential diagnoses, and it is proposed to continue carrying out related studies to standardize a definitive management that impacts the quality of life of the patient and reduces the risk of death.

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