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Case Report

Cholangitis Secondary to Lemmel Syndrome: Case Report

Abstract

Background: Acute cholangitis is a life threatening condition with multiple possible origins; one of this is the distal intermittent obstruction of biliary tree by a duodenum yuxtapapillar diverticulum, an infrequent condition known as Lemmel syndrome.

Case: An 83-year-old man come to emergency room complaining for intense abdominal pain and fever. Pathological background included cholecystectomy 4 years ago and right inguinal hernia repair 2 years before. He presents with epigastric, moderate intensity pain 4 days before, developing general discomfort, anorexia, asthenia, adinamia and nausea. Last 24 hours presents fever of 39.5° and jaundice. At physical exam with Glasgow 12, jaundice, dehydrated, tachycardia of 95 beats per minute, tachypnea 25 breaths per minute, abdominal pain located in epigastrium and right hypochondria, Murphy +, extremities with delayed capillary fulfill. Laboratories report hemoglobin 14.7 mg/dL, leucocytes 22000, neutrophils 95%, platelets 18500, total bilirubin 17 mg/dL, direct bilirubin 13.2 mg/dL. Ultrasound reports gallbladder absence, with intrahepatic biliary ducts dilated 7mm, and common bile duct 9 mm without intraluminal content. Medical treatment including metronidazole plus imipenem were initiated with good results and cholangitis resolution. ERCP was performed and this study reports a yuxtapapillar diverticulum type 1, a sphincterotomy was completed with clear biliary liquid evacuated and with posterior 10 French x 10 cm endoprothesis placement. Two days after that the patient was discharged but after one week he returned by mild cholangitis. Colangio-pancreatic magnetic resonance report similar findings that in ERCP, with yuxtapapillar duodenal diverticulum. By clinical presentation and evolution Lemmel syndrome was diagnosed and conservative management with ERCP and sphincterotomy completed by the good outcomes, with patient discharge uneventfully 4 days later.

Conclusions: The intermittent obstruction of biliary tree by a duodenal diverticulum is a rare condition that must be suspected in cases of repetitive cholangitis and no evidence of choledocholithiasis, confirmed by ERCP and discarding another anatomical abnormality by magnetic colangio-pancreatic magnetic resonance.

Introduction

Lemmel Syndrome was first described in 1934, and defined as an obstruction of biliary tree secondary to duodenal diverticulum. The incidence of duodenal diverticulum is about 1-10% and until 27% in other series, and is the second more frequent site for diverticula location only after colon. From the peri-ampullar location near 10% could be symptomatic along live, requiring endoscopic or surgical management to resolve the biliary obstruction, being important to identify it as a cause of intermittent jaundice in absence of choledocholithiasis [1,2].

Case

An 83-year-old man come to emergency room complaining for intense abdominal pain and fever. Pathological background

included cholecystectomy 4 years ago and right inguinal hernia repair 2 years before.

He began with epigastric moderate intensity pain 4 days before, developing general discomfort, anorexia, asthenia, adinamia and nausea. Last 24 hours' present fever of 39.5° and jaundice.

At physical exam with Glasgow 12 points, jaundice, dehydrated, tachycardia 95 beats per minute, tachypnea 25 breaths per minute, abdominal pain located in epigastrium and right hypochondria, Murphy +, extremities with delayed capillary fulfill. Laboratories report hemoglobin 14.7 mg/dL, leucocytes 22000, neutrophils 95%, platelets 18500, Glucose 207 mg/dL, Creatinine 1.8 mg/dL, Na 120 mEq/L, Cl 94 mEq/L, K 3 mEq/L, total bilirubin 17 mg/dL, direct bilirubin 13.2 mg/dL, ALT 118 UI/L, AST 160 UI/L, FA 192 UI/L, TP 18 seconds, TPT 30.1

seconds, INR 1.4, gas analysis: pH 7.45, Pco₂ 62 mmHg Po₂, 62 mmHg, hco₃ 16 mmol/L, Baeef -6.2 mmol/L. Ultrasound report gallbladder absence, with intrahepatic biliary ducts dilated 7mm, and common biliary duct 9 mm without intraluminal content. Medical treatment including metronidazole plus imipenem were initiated with good results and cholangitis resolution. ERCP was performed and this study reports a yuxtapapillar diverticulum type 1, with cholangiography showing a dilated biliary tree without defects, a sphincterotomy was completed with clear biliary liquid evacuated and with posterior 10 French x 10 cm endoprosthesis placement (Figure 1). Two days after that the patient was discharged but after one week he returned by mild cholangitis. Colangio-pancreatic magnetic resonance was requested and report similar findings that in ERCP, with yuxtapapillar duodenal diverticulum (Figures 2,3). By clinical presentation and evolution Lemmel syndrome was diagnosed and conservative management with ERCP and sphincterotomy completed by the good outcomes associated, with patient discharge uneventfully 4 days later.

Discussion

Duodenal diverticulum incidence is estimated between 1 to 27% in general population depending on the consulted series. It is usually founded incidentally. According to the anatomical classification after colangio-pancreatic magnetic resonance are divided in four types, with the yuxtapapillar or periampullar being the most frequent in 75% of cases [3]. Related symptoms include chronic abdominal pain, gastrointestinal bleeding, diverticulitis, perforation, intestinal obstruction and intermittent jaundice in absence of choledocholithiasis

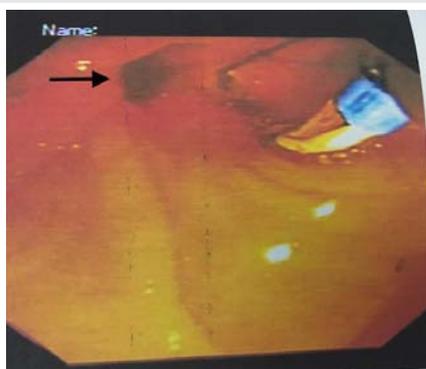


Figure 1: Yuxtapapillar diverticulum (black arrow) at less than 3 cm from the Vater ampulla with stent.

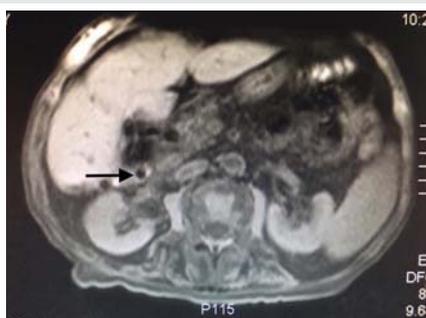


Figure 2: Cholangioresonance with evidence of duodenal diverticulum lumen (black arrow).



Figure 3: Cholangiography by magnetic resonance with evidence of duodenal diverticulum (black arrow)

known as Lemmel syndrome, and less than 1% require surgical treatment [4].

In the presented case the diagnosis was suspected only after the second cholangitis event by the low incidence of this pathology and the possibility of a transient choledocholithiasis, the most frequent origin of common biliary obstruction. Unfortunately, although the correct medical and endoscopic management indicated in this case, with the endoprosthesis placement in common biliary duct, the patient presents another episode of cholangitis, requiring more imaging exams to discard another associated alteration, and once discarded, endoscopic management was selected again with good outcomes this time.

The pathophysiology is explained by some different theories including diverticulitis or mechanical irritation of periampullar diverticula that may cause inflammation of the ampulla that would lead to fibrosis. Another theory is that diverticula itself may cause dysfunction of sphincter of Oddi by anatomical modifications of angles and sphincters, in fact distal common biliary duct or ampulla can be compressed mechanically leading to jaundice, sometimes intermittent, and in other cases favoring the increase on incidence of choledocholithiasis by colonization and overgrowth of β -glucuronidase producing bacteria, which in turn lead to deconjugation of bilirubin glucuronides and results in precipitation of calcium bilirrubinate gallstones [2].

The diagnosis usually is by direct visualization of duodenal diverticula by endoscopic ultrasound and endoscopic retrograde cholangiopancreatography (ERCP), and confirmed by colangio-pancreatic magnetic resonance as the gold standard, describing in this and CT scan images a thin-walled cavity lesions situated on the medial wall of the duodenum 2nd portion that typically contain gas [2,3].

The preferred treatment and with the better results and reduced morbidity is the ERCP with sphincterotomy and endoprosthesis placement, with cannulation rates of 94.9% in some series, and complications like bleeding or perforation in 7.8% [1,5]. There is a consensus that elective surgical treatment of asymptomatic diverticulum is not justified but in case of require this approach the diverticulectomy or biliodigestive anastomosis would be the surgical options [1,2,4].



The Lemmel syndrome is a rare condition that must be suspected in cases of intermittent obstruction of biliary tree with or without cholangitis and no evidence of choledocholithiasis, confirmed by ERCP and discarding another anatomical abnormalities by colangio-pancreatic magnetic resonance to offer the correct and best management for each specific case. Ignoring this possibility could lead to repetitive jaundice and in some cases cholangitis, with the high morbidity and mortality associated.

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