Chirurgia. eCollection 2024 August
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http://dx.doi.org/10.21614/chirurgia.119.eC.2998

Case Report: An Unreported Presentation of Lemmel Syndrome Highlighting Diagnostic Challenges of Duodenal Masses

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Rezumat

Prezentare de caz: provocările diagnostice ale formațiunilor tumorale duodenale - sindromul Lemmel

Prezentăm cazul unui pacient în vârstă de 67 de ani, cunoscut cu bypass gastric, recent diagnosticat cu o formațiune tumorală duodenală de 6 centimetri, stenozantă la nivelul căii biliare principale. În ciuda multiplelor tentative, abordul endosopic al formațiunii tumorale nu s-a putut realiza, fiind necesară intervenția chirurgicală. În timpul explorării chirurgicale, s-a constatat prezența unui diverticul duodenal locuit în totalitate de calculi, care a produs obstrucția căii biliare - o manifestare a sindromului Lemmel. Această afecțiune rară se caracterizează prin icter obstructiv în absența coledocolitiazei sau a tumorilor şi este secundară dilatării unui diverticul periampular. Deşi abordul acestei afecțiuni este, de regulă, endoscopic, în cazul de față, accesul endoscopic cu scop diagnostic şi terapeutic a fost complicat de antecedentele chirurgicale ale pacientului (bypass gastric). Această prezentare de caz evidențiază provocările impuse de o patologie rară suprapusă pe fondul unei anatomii modificate chirurgical, situație întâlnită din ce în ce mai frecvent în practica chirurgicală zilnică. În astfel de situații, laparotomia exploratorie continuă să joace un rol semnificativ.

Cuvinte cheie: sindromul Lemmel, formațiune tumorală duodenală, stenoză a ductului biliar, bypass gastric, anatomie modificată chirurgical

Received: 30.05.2024
Accepted: 05.08.2024

Abstract

We present a case involving a 67-year-old patient with a medical

history of gastric bypass who was recently diagnosed with a 6-centimeter duodenal mass causing biliary duct stenosis. Despite our best efforts, we were unable to access this tumor endoscopically, necessitating surgical intervention. During the surgical exploration, we discovered a duodenal diverticulum filled with stones, leading to the obstruction of the biliary duct—a manifestation of Lemmel syndrome. This rare condition is characterized by obstructive jaundice in the absence of choledocholithiasis or tumors and is secondary to dilatation of peri-ampullary diverticula. While it is typically managed through endoscopy, our diagnostic and therapeutic approach was complicated by the patient's history of bariatric surgery (gastric bypass), making endoscopic access impossible despite our multiple attempts. This case report sheds light on the challenges posed by the concurrence of a rare pathology and surgically modified anatomy, which is increasingly encountered in daily surgical practice. In such situations, exploratory surgery continues to play a significant role.

Key words: Lemmel's syndrome, duodenal mass, Bile duct stenosis, gastric bypass, surgically altered anatomy

Introduction

Obstructive jaundice is a common symptom, often caused by choledocholithiasis or peri-ampullary tumors. Peri-ampullary diverticula are extraluminal outpouchings of the duodenal mucosa that develop within 2 to 3 cm from the ampulla of Vater. These diverticula are usually asymptomatic and are typically discovered incidentally. However, obstructive jaundice can develop, in the absence of choledocholithiasis or tumor, due to distended peri-ampullary diverticula (1). This condition is mostly managed endoscopically (1-5). However, when this uncommon condition occurs in a patient with surgically altered anatomy, the management becomes more challenging.

Case Report

A 67-year-old patient was admitted to our surgical department following diagnosis of a 6-centimeter duodenal tumor. The patient's medical history was significant for a hysterectomy and a gastric bypass procedure, which had to be converted from laparoscopy to laparotomy due to major peritoneal adhesions.

Due to recurrent abdominal pain, a CT scan was performed which revealed a duodenal mass with a hypo-enhancing center. This mass resulted in distal bile duct stenosis and mild intra-hepatic biliary tract dilation (*Figs. 1, 2*). She was subsequently referred to the gastro-intestinal care unit for further evaluation by blood tests and endoscopy. There were no significant abnormalities in the blood analysis except a mildly elevated CEA level at 8.7 µg/L. Endoscopic assessment (ballon enteroscopy) was unsuccessfull, the gastroenterologist was unable to ascend the biliopancreatic loop to the duodenum. Therefore, the patient was transferred to the surgical unit for further investigations.

Due to her surgical history and the presence of intra-abdominal adhesions, laparoscopy was deemed unsafe. Consequently, a small laparotomy was performed to assess the lesion and establish a temporary gastrostomy in the excluded stomach to facilitate endoscopy. Intra-operatively, the mass was identified as a hard but, unexpectedly, very mobile intra-duodenal mass. There was no evidence of invasion of the adjacent structures. Subsequently, a duodenotomy was performed, revealing the presence of a peri-ampullary diverticulum containing stones that likely caused the bile duct stenosis (*Figs. 3, 4*).

The stones were extracted, followed by a cholecystectomy and the placement of a transcystic drain through the bile duct to safeguard it. The diverticulum was closed using non-

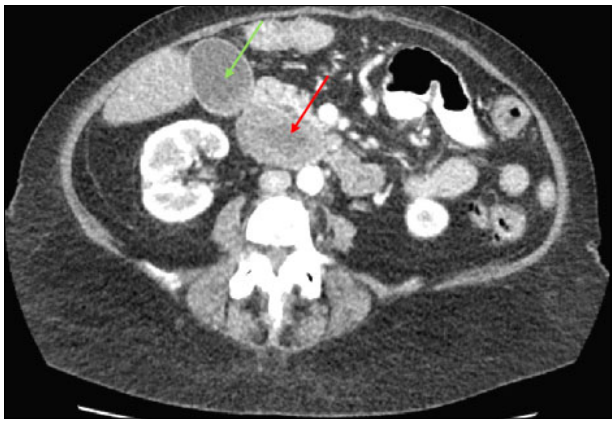


Figure 1. Transversal CT-scan image of the duodenal mass. Duodenal mass (red arrow). Gallbladder (green arrow)



Figure 2. Coronal CT-scan image of the duodenal mass. Duodenal mass (red arrow); gallbladder (green arrow). Dilated bile duct (yellow arrow)

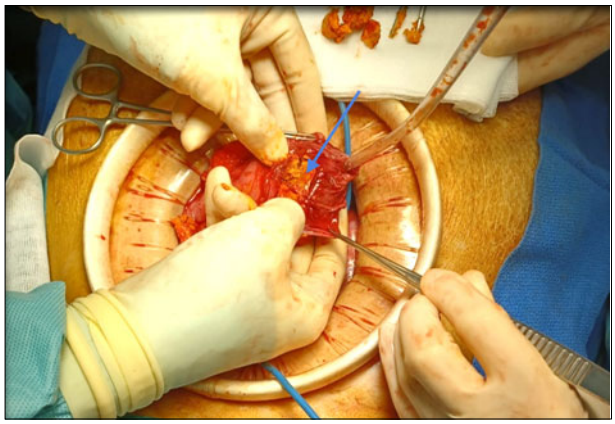


Figure 3. Per-operative photograph of the diverticulum filled with lithiasis (blue arrow)

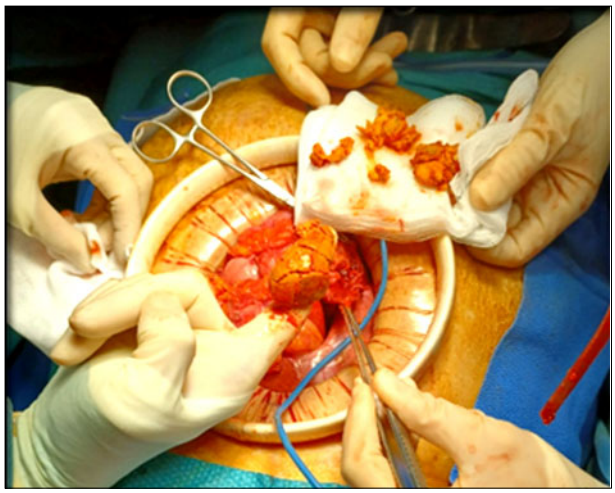


Figure 4. Extracted lithiasis of the diverticulum

resorbable sutures. The duodenotomy was closed using absorbable sutures, and a drain was placed in contact with the suture line. Unfortunately, on the ninth day post-operation, the patient developed hemorrhagic shock due to bleeding from a duodenal ulcer unrelated to the previous suture site. Surgical intervention was necessary for hemostasis. The patient subsequently experienced a swift recovery, and the trans-cystic tube was removed eight weeks after the operation following duodenal frame opacification.

At follow up six months later, the patient shows no signs of recurrence (*Figs. 5, 6*).

Discussion

Lemmel's syndrome is characterized by a peri-ampullary diverticulum causing compression of the pancreatic or common bile duct, leading to obstructive jaundice. While duodenal diverticula are the second most common after colonic diverticula, they are classicaly asymptomatic and discoverd incidentally (1-4). Nevertheless, periampullary diverticula can result in pancreaticobiliary obstruction and obstructive jaundice. Seldom, they can cause

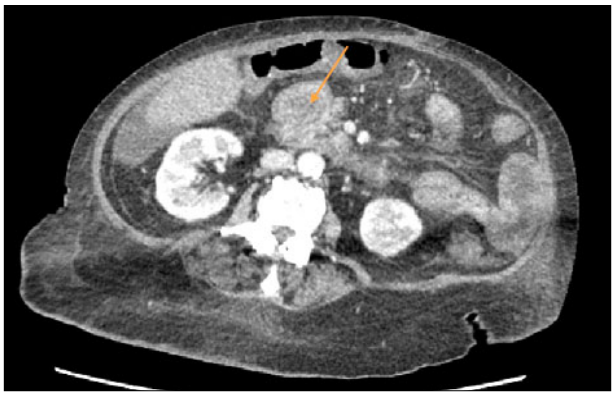


Figure 5. 6-month follow-up CT scan. Duodenum (Orange arrow)



Figure 6. 6-month follow-up Coronal CT Scan. Duodenum (Orange arrow)

intestinal obstruction, hemorrhage, fistula or perforation (3). Lemmel syndrome can be diagnosed radiologically, for example with the use of CT imaging, or by direct visualization endoscopically. Magnetic resonance cholangiopancreatography (MRCP) is also a useful imaging modality in the diagnosis of biliary tract pathology and in characterizing periampullary diverticula (13-14). In our case, a CT-scan showed a rapidly growing mass with a hypo-enhanced center in the context of a patient with an elevated CEA, raising concerns that she was at risk of a malignant tumor. Therefore, we chose not to delay the endoscopic visualization in waiting for an MRI.

Endoscopic extraction of material and sphincterotomy constitute the standard treatment, sometimes involving bile duct stent placement (4-5). Surgical options may be considered for recurrent cases, allowing diverticulectomy or biliary derivation (6 -7 -10).

When a peri-ampullar mass is discovered, it is necessary to visualize the mass and obtain histopathological sample in order to make a diagnosis. In the majority of patients, this can be achieved endoscopically (8). Our patient presented with both diagnostic and therapeutic challenges due to her surgical history. While most patients can be diagnosed endoscopically, those with altered anatomy present difficulties accessing duodenum and Ampulla of Vater. There is currently no established method for optimal ampulla access in these cases (9). Balloon and spiral enteroscopy, along with hybrid techniques combining ultrasound or laparoscopy with conventional endoscopy, have been explored. A meta-analysis and review of the litterature by Gkolfakis et al. demonstrates higher efficacy, but increased adverse events, with laparoscopy and ultrasound - ERCP. Currently, there is a lack in randomized studies to assess the best approach in the anatomically altered patient, though this can be explained by the technical difficulties and low incidence of the pathology (9-10).

Our case report highlights the value of surgical exploration in hybrid methods like laparoscopic-ERCP (LA-ERCP), enabling macroscopic assessment and exploration of the abdominal cavity. Tonnesen et al.'s study comparing laparoscopy-assisted and balloon enteroscopy-assisted ERCP also emphasizes the success of both methods, highlighting the potential benefits of LA-ERCP especially when procedures such as cholecystectomy or exploratory laparoscopy are indicated (11).

The surgical approach to Lemmel syndrome is challenging and there is no consensus, with

most described cases in the literature being case reports (12). Furthermore, the association between this syndrome and surgically altered anatomy is notably absent from the literature. Diverticulectomy is hindered by the challenging posterior diverticulum location, particularly in cases with inflammation or extensive adhesions, as seen in our patient. Biliary derivation, while an option, carries high morbidity (13,15). While the literature reports surgical approach such as biliary derivation and diverticulectomy in case of endoscopic failure, we did not obtain consent for those high-risk procedures. Therefore, in our patient's case, we opted for diverticulum drainage and closure, in addition to cholecystectomy to decrease the risk of future lithiasis entrapment. If the patient shows signs of recurrence in the future it will then still be possible to propose a more radical surgical approach in a more specialized environment.

Conclusion

Lemmel syndrome, a rare condition, should be considered in cases of non-stone-induced jaundice, particularly when duodenal diverticula history exists. The coexistence of surgically altered anatomy further complicates diagnosis and management. This case emphasizes the continued role of exploratory surgery in assessing and treating abdominal masses, especially in cases where endoscopy is not feasible which can easily occur in patients with surgically impaired anatomy.

Statement on Funding Sources and Conflicts of Interest

No financial support and no other potential conflicts of interest relevant to this article were reported.

Informed Consent

Written informed consent was obtained from the patient for publication.

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