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Case Report

Lemmel syndrome: a rare condition causing cholangitis due to duodenal diverticulum

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ABSTRACT

Lemmel syndrome, rare condition occurs when there is one or more diverticulum causes cholangitis and obstructive jaundice due to compression of common bile duct (CBD) by mechanical obstruction. And are commonly recognized as an incidental finding at cross-sectional imaging. There may be also added pathophysiologic mechanisms that also contribute to the process to the development of this syndrome like duodenal diverticula leading to sphincter of Oddi dysfunction and pressure compression of CBD by diverticula. Inflammation of these duodenal diverticula is very rare. Patients undergoing routine gastrointestinal tract (upper GI) evaluation, duodenal diverticulum presents up to 27% of patients mostly periampullary diverticulum. There may be multiple diverticula leading to compression of CBD causing cholangitis.

Keywords: Lemmel syndrome, Sphincter of Oddi dysfunction, Periampullary diverticulum

INTRODUCTION

Lemmel syndrome was first defined by Lemmel in 1934 as a diverticulum of the periampullary duodenum causing obstructive jaundice in the absence of choledocholithiasis or neoplasm**.** Mr Lemmel firstly described Lemmel syndrome in 1934 as a rare cause of obstructive jaundice. Generally, this type of duodenal diverticulum has a wide mouth orifice and for this reason bezoar of enterolith are commonly pushed to duodenum without sequelae. But by time with multiple time infection of inflammation narrows the mouth of diverticulum and thus bezoar or enterolith trapped in duodenal diverticulum causing mass effect of CBD resulting in cholangitis and obstructive jaundice.

We reported this case a 52-year-old man presented with pain in right hypochondrium with features of cholangitis, sepsis and acute kidney injury (AKI). His initial lab were concerning for infection with cholangitis, AKI and abnormalities of hepatobiliary tree. On Further investigation a CECT abdomen was obtained which shows two large diverticula close to periampullary region compressing CBD from both sides.

CASE REPORT

A 52-year-old man who is a known smoker and has history of social ethanol use was admitted with fever for 3 days, icterus for 2 days and right hypochondriac pain. He also showed local muscular guard in right hypochondriac region. At the time of admission patient showed evidence of breathlessness with oliguria. Patient had evidence of leucocytosis with total leukocyte count (TLC)- 18600, differential count (DLC)= neutrophil-65%, lymohocyte-25%, monocyte- 9%, eosinophil- 1%, basophil- 0%, with obstructive jaundice with total bilirubin of 9.0 mg/dl, conjugated billirubin- 6.5 mg/dl, SGOT- 111, SGPT- 65, alkaline phosphate- 480 units/l and acute kidney injury with urea- 234 mmol/l, creatinine- 4.5 mg/dl and urine volume was less than 400 ml/day. Patient also had

evidence of ARDS with B/l chest infiltrate, with normal echocardiography. His treatment was initiated with IV fluid, antibiotics- piperacillin and tazobactam with doxycycline along with supportive management. Within 48 hours of treatment patient showed remarkable improvement with TLC- 9750, DLC= neutrophil- 50%, lymphocyte- 45%, monocyte- 4%, eosinophil- 1%, basophil- 0% and urea- 55 mmol/l, creatinine- 1.2 mg/dl, LFT= total bilirubin- 2.3 µmol/l, conjugate bilirubin- 1.3 µmol/l, SGOT- 89, SGPT- 105, alkaline phosphate- 449 units/l and chest became clear and disappearance of lung infiltrate. He fulfilled Charcot's triad for cholangitis. In our endeavour to look for the cause we found in MRCP showed- (1) a round soft tissue lesion abutting over terminal end of CBD and adherent to duodenal wall, and (2) multiple simple cystic lesions in liver.

He was thereafter referred to a radiologist for CECT w/a showed- 1) multiple duodenal diverticuli; (2) hepatomegaly with multiple cysts, (3) nephrolithiasis-right and simple cortical cysts of left kidney; (4) small adrenal adenoma on the left side. Meanwhile, around four cases were reported in Europe recently between 2019 and 2020. In these cases, the presentation was mainly acute recurrent abdominal pain, which required a visit to emergency services. Diagnosis of Lemmel syndrome was made based on CT findings, which showed a typical PAD with associated infection.

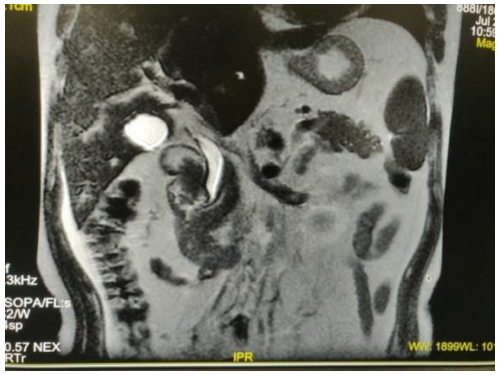

Figure 1: MRCP shows CBD narrowing at terminal part.

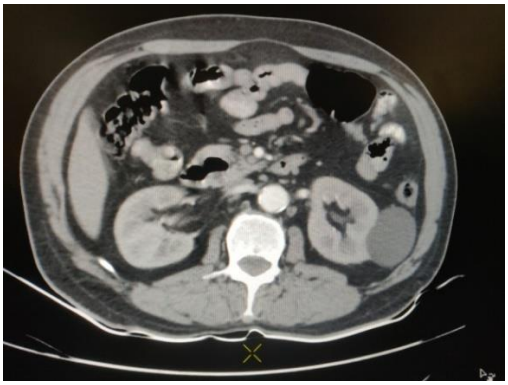

Figure 2: CECT whole abdomen showing two diverticula compressing lower end of CBD.

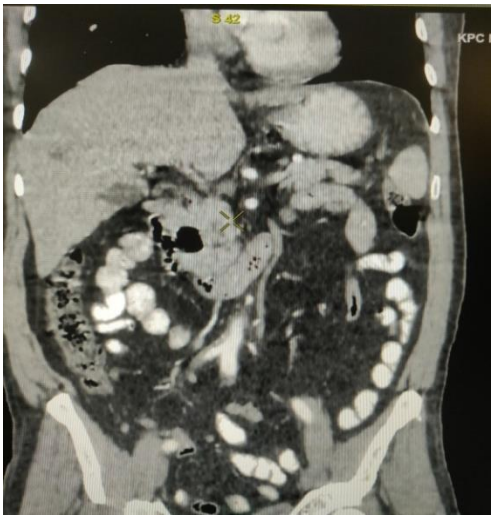

Figure 3: CECT whole abdomen showing diverticulum.

DISCUSSION

This syndrome should be considered in the absence of choledocholithiasis or other more common causes of obstructive jaundice. Periampullary duodenal diverticula typically occur along the medial aspect of the second or third part of the duodenum.[1] They are rarely symptomatic.[2] To date, very few cases have been published and fully investigated.[3-5] Lemmel syndrome resulting from papillitis chronica fibrosa or sphincter of Oddi dysfunction can be successfully treated with endoscopic sphincterotomy to release the biliary obstruction.[6] Lemmel syndrome was first defined by Lemmel in 1934 as a diverticulum of the periampullary duodenum causing obstructive jaundice in the absence of choledocholithiasis or neoplasm.[7] Duodenal diverticula specifically periampullary duodenal diverticulum generally occurs along medial side of second and third part of duodenum adjacent to ampulla of vater and the very rarely become symptomatic. Patients generally improve with administration of antibiotics, placement of plastic biliary stent by ERCP, and drainage of the obstruction, respectively.[8-10] If at all symptomatic and it leads to complication less than 5% of cases including cholangitis, pancreatitis, bleeding, perforation, bezoar and very rarely obstructive jaundice. Most of the cases of Lemmel syndrome presents to us with acute cholangitis, pain abdomen and obstructive jaundice and symptoms can be intermittent. Our patient presented with features of cholangitis with elevated TLC, ESR, CRP, and deranged LFT and RFT though blood culture was negative. We made the diagnosis confidently with help of similar clinically history, MRCP, CECT of whole abdomen.

CONCLUSION

Cholangitis is a common problem encountered in routine clinical practice. However this case we present a rare cause of duodenal diverticulum causing cholangitis. The

admixture of clinical presentation which suggestive of episode of cholangitis and imaging findings lean towards most likely diagnosis, Lemmel syndrome which is obstructive jaundice due to an impacted duodenal diverticulum.

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