

Rare Cause of Sphincter of Oddi Dysfunction and Double Duct Sign

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Abstract

Double duct sign on imaging is defined as dilated common bile duct and pancreatic duct till ampulla and is usually associated with an ampullary obstruction likely periampullary malignancy. Sphincter of Oddi dysfunction is an uncommon cause of double duct sign. In the present case, we are discussing a rare cause of Sphincter of Oddi dysfunction with double duct sign.

Keywords

- ▶ Double Duct Sign
- ▶ Sphincter of Oddi Dysfunction
- ▶ Endoscopic Ultrasound
- ▶ Morphine
- ▶ Opium

Introduction

Dilatation of the common bile duct (CBD) along with pancreatic duct (PD) is an important radiological sign suggesting an obstruction at the distal portion of both the CBD and PD.¹ The presence of a double duct sign should ensure a careful search for underlying etiology. Though periampullary malignancy is the most common cause, benign diseases such as chronic pancreatitis and sphincter of Oddi dysfunction (SOD) can also lead to double duct sign. SOD is due to a functional obstruction at papilla and high index of suspicion is required for diagnosing this condition after ruling out other structural causes. We hereby report a rare cause of SOD giving rise to a double duct sign.²

Case Presentation

A 57-year-old male businessman presented with complaints of episodes of noncolicky epigastric pain, without any history of fever, jaundice, or weight loss. Each episode used to last 5 to 6 hours with either spontaneous improvement or with parenteral analgesics. Clinical examination was unremarkable without any icterus or abdominal lump. Blood inves-

tigations revealed elevated liver enzymes that were suggestive of cholestatic/obstructive pattern. Serum bilirubin value was normal (1.2 mg/dL), and serum alkaline phosphatase, gamma glutamyl transpeptidase, serum glutamic pyruvic transaminase, and serum glutamic-oxaloacetic transaminase values were 579 IU/L, 2,309 IU/L, 249 IU/L, and 186 IU/L, respectively. Ca 19-9 was elevated to two times the upper limit of normal. Viral markers hepatitis B surface antigen, anti-hepatitis C virus, immunoglobulin M (IgM) anti-hepatitis A virus, IgM anti-hepatitis E virus were negative. Ultrasound (US) abdomen revealed dilated CBD measuring 12 mm till the lower end without any stone or mass lesions, and distended gallbladder with sludge. Triple-phase computed tomography revealed dilated CBD and PD measuring 12 and 8 mm, respectively, till lower end without any mass lesion (▶ Fig. 1). Magnetic resonance cholangiopancreatography showed dilatation of bilobar intrahepatic biliary radicles, bile duct, and PD without any mass lesion (▶ Fig. 2). Endoscopic US (EUS) (▶ Video 1) was performed that showed dilated CBD and PD till ampulla without any mass lesion or stone, smooth symmetrical narrowing of bile duct and PD at papilla, symmetrically thickened ampullary without any mass lesion (▶ Fig. 3), and normal pancreatic

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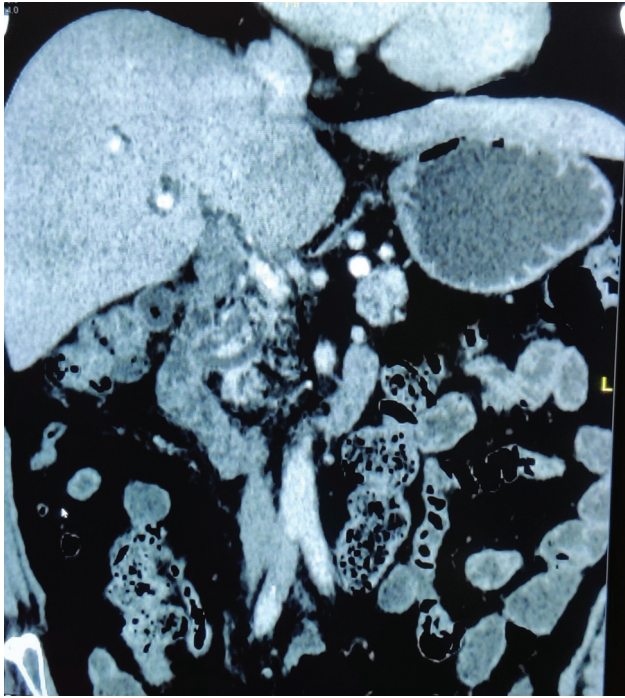


Fig. 1 Computed tomography findings showing double duct sign.

parenchyma without any evidence of chronic pancreatitis. Based on clinical, biochemical and imaging findings, differential diagnoses of ampullary neoplasm and SOD were considered. Human immunodeficiency virus serology was negative. Clinical history was revisited, which revealed significant opium intake for the last 20 years for recreational purpose. Based on clinical findings, imaging, and EUS, patient was suspected to have SOD, and decision to perform endo-

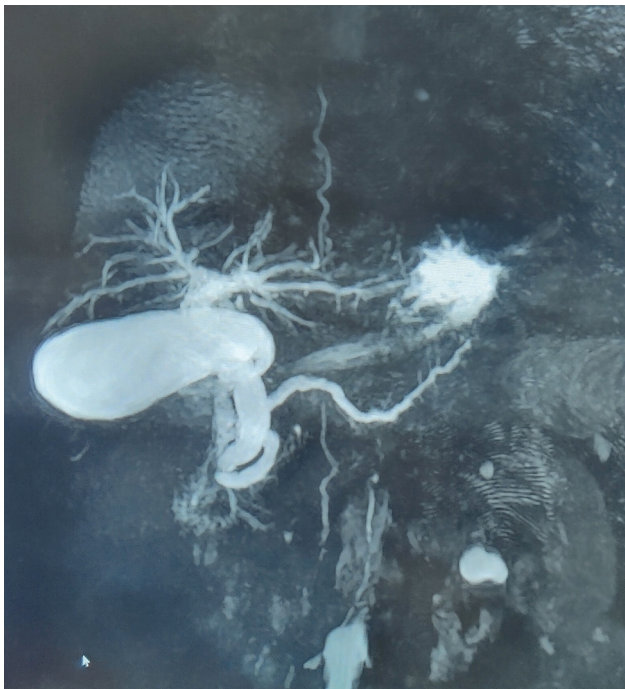


Fig. 2 Magnetic resonance cholangiopancreatography findings showing double duct sign.

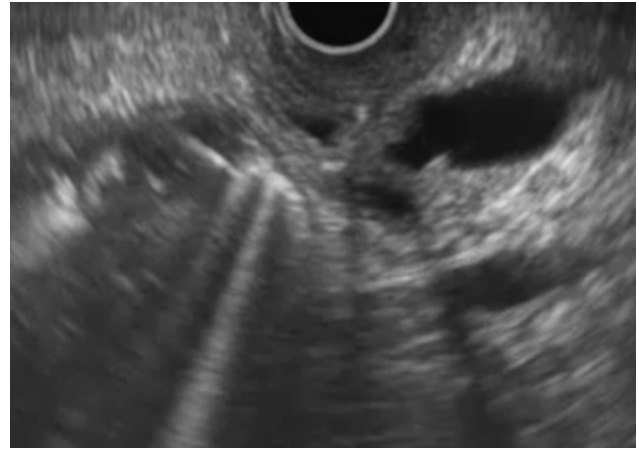


Fig. 3 Endoscopic ultrasound showing double duct in the absence of mass lesion and smooth tapering of ducts.

scopic retrograde cholangiopancreatography (ERCP) and sphincterotomy was taken after explaining the procedure details and possible complications to the patient. Side-view endoscopy revealed stenotic opening (→**Fig. 4**) at the papilla, which could be cannulated with maneuvering. Endoscopic papillotomy was performed that also revealed thick ampullary muscle. (→**Video 1**) Ampullary biopsy was taken which did not reveal dysplasia or malignancy (→**Fig. 5**).

Patient was discharged after 24 hours of the procedure and was symptom free after 3 months of procedure. Repeat investigations revealed normalization of liver function test and Ca 19-9. Patient was advised to stop the opium intake and was advised for enrollment in a de-addiction clinic. He is under continuous follow-up to rule out the possibility of occult biliary malignancy.

Discussion

Dilated pancreatic and common bile duct (double duct sign) commonly occur due to periampullary malignancy. Chronic pancreatitis and SOD are possible benign differentials that should be considered in patients without mass lesion on imaging.

SOD refers to an abnormality of Sphincter of Oddi contractility causing obstruction to the flow of bile or pancreatic juices.³ Sphincter of Oddi manometry had been identified as a gold standard for diagnosis of SOD; however, it is invasive and associated with morbidity and hence has been largely abandoned. Patients with type 1 SOD should be treated with endoscopic sphincterotomy without manometry.⁴

In our case, SOD was considered as likely possibility due to significant history of opium intake and absence of mass lesion or chronic pancreatitis on imaging. EUS is an important investigation to diagnose this condition by ruling out other etiologies such as neoplastic lesions or chronic pancreatitis.⁵ Other EUS findings supporting the diagnosis were smooth symmetrical narrowing of ducts at ampulla and symmetrically thickened ampullary muscles. ERCP findings of circumferential thickening of ampullary muscle also supported the diagnosis of SOD.

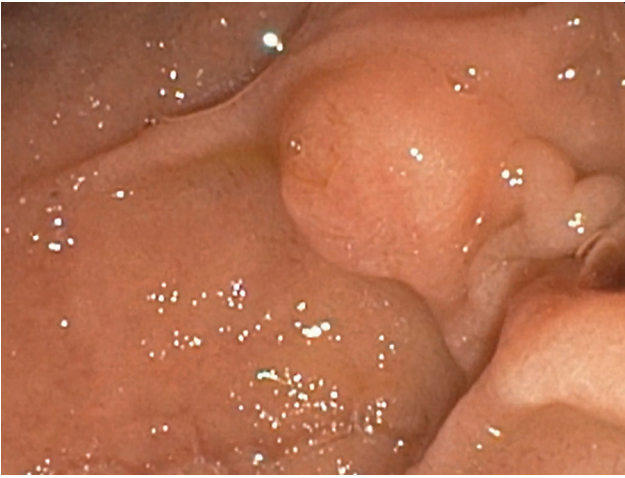


Fig. 4 Side-view endoscopy showing stenotic and thickened papilla.

Morphine and its derivatives can increase sphincter tone and result in SOD.^{6,7} Morphine use can cause a rise in basal pressure of sphincter of Oddi along with a rise in amplitude and frequency of phasic contractions. CBD pressures have also been shown to increase after morphine intake. Hence, opium deaddiction forms a cornerstone in the management of opium-induced SOD as it helps to reverse the pharmacological effects of morphine. However, prolonged opium use can also lead to bulky tumorous changes in ampulla with ulceration.⁸ In such

cases, long-term improvement has been seen after endoscopic sphincterotomy.^{7,9} Therefore, a history of opium abuse must be sought in every patient presenting with double duct sign without evidence of a mass lesion causing ampullary obstruction.

Conclusion

Opium intake can lead to SOD and dilated PD and bile duct. EUS is an important diagnostic modality in such cases to rule out small ampullary tumors. Endoscopic sphincterotomy can lead to long-term relief of symptoms in patients with SOD.

Video 1

0:00-0:34: Endoscopic ultrasound examination revealed dilated bile duct and pancreatic duct with smooth narrowing at ampulla, symmetrical thickening of ampullary muscle without any mass lesion.
0:34-1:23: Cannulation of stenotic ampulla.
1:23- 2:18: Endoscopic sphincterotomy with balloon sweep of bile duct. Online content including video sequences viewable at: <https://www.thieme-connect.com/products/ejournals/html/10.1055/s-0044-1785223>.

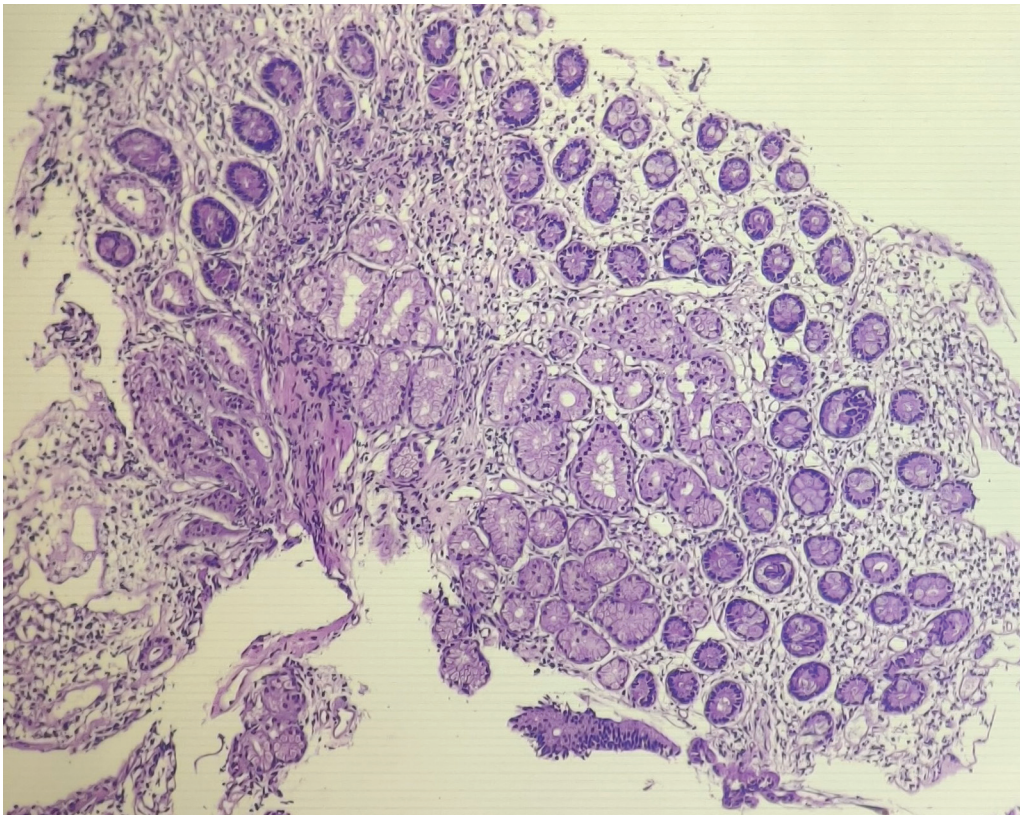


Fig. 5 Ampullary biopsy—negative for malignancy or granuloma.

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Conflict of Interest

None declared.

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