Published online: 2019-09-26

Case Report

An uncommon evaluation in recurrent acute pancreatitis

Mallikarjun Patil, Keyur A. Sheth

Departments of Gastroenterology, St. John's Medical College, Bengaluru, Karnataka, India

Abstract

Recurrent acute pancreatitis can be caused due to mechanical, metabolic, anatomical abnormalities and other miscellaneous causes. Among the anatomical causes, choledochoceles are rare entities that result in recurrent acute pancreatitis. Choledochoceles are different in morphological and clinical features as compared to other choledochal cysts. We present a case of 21-year-old female with recurrent episodes of acute pancreatitis due to choledochocele as etiology. She was managed with endoscopic sphincterotomy and was asymptomatic for last 24 months.

Key words

Cholangiography, choledochocele, pancreatitis, recurrent, sphincterotomy

Introduction

Acute pancreatitis is described by the presence of inflammation of the pancreas, which is otherwise normal. Etiology of pancreas if remains untreated, it results into recurrent acute pancreatitis. Recurrent acute pancreatitis is characterized by two or more episodes of acute pancreatitis with complete or near complete improvement of clinical features of pancreatitis between episodes. Choledochoceles are identified as cystic dilatations of the intraduodenal portion of the common bile duct (CBD); however, they stand apart from other usual choledochal cyst by their different morphological and clinical features. We present a case of recurrent acute pancreatitis with choledochocele as etiology.

Case Report

A 21-year-old female presented to us with complains of sudden onset abdominal pain for the last 48 h. The abdominal pain was located in epigastric region, severe in intensity, radiating to back, aggravated by food and relieved by sitting in the forward position. She had a history of similar two episodes of pain in past 2 years with this being the third episode. There was no history of jaundice, itching, fever, alcohol intake or trauma. She was not taking any other medications at presentation. There was no family history of similar pain or pancreatitis. Physical examination was unremarkable except tenderness in the epigastric area. Laboratory parameters revealed normal hemogram and liver biochemistry. Serum level of amylase was 786 (normal range 25–115 U/L) and lipase was 1212 (normal range 114–286 U/L), which were elevated more than 3 times the upper limit of normal. Serum level of ionized calcium was 1.20 mEq/L (normal range 1.16–1.32) and triglycerides was 127 mg/dl (range <150 mg/dl), which were normal. Transabdomen Ultrasound showed a bulky pancreas with no pancreatic duct dilatation or calcifications and normal biliary anatomy. Endoscopic ultrasound (EUS) [Figure 1] demonstrated rounded protrusion of intramural portion of distal CBD proximal to the ampullary orifice. The lesion was separate from duodenal muscle layers and contained sludge inside. EUS also showed normal proximal CBD and biliary radicles. The gall bladder was distended and anechoic with wall thickness of 2 mm. The pancreas was bulky with duct measuring 2 mm in head and 1 mm in body. Endoscopic retrograde cholangiography (ERC) [Figures 2 and 3] shows retention of contrast medium within cystic dilation at the terminal CBD with normal proximal CBD and biliary radicles. There was no intraluminal filling defects. With the help of 0.025 inch guide wire and sphincterotome, biliary sphincterotomy [Figure 4] was performed. After wide biliary

Access this article online

Website: www.jdeonline.in

Quick Response Code

DOI: 10.4103/0976-5042.147504

Address for correspondence:
Dr. Mallikarjun Patil, Department of Gastroenterology, St. John's Medical College, Bengaluru - 560 034, Karnataka, India. E-mail: drmalli_arjun@yahoo.co.in

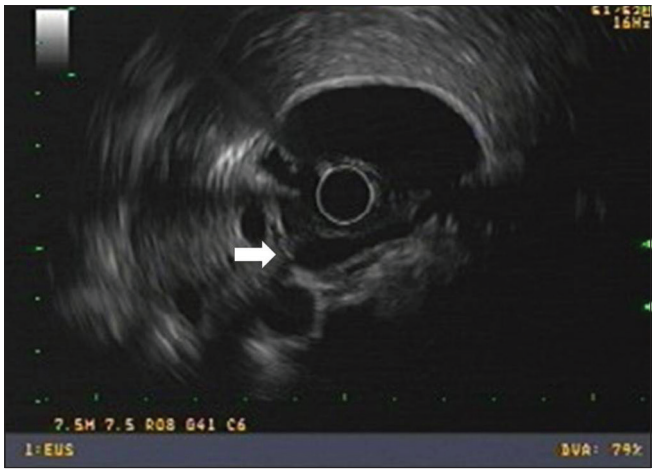

Figure 1: Endoscopic ultrasound image showing protrusion of intramural common bile duct

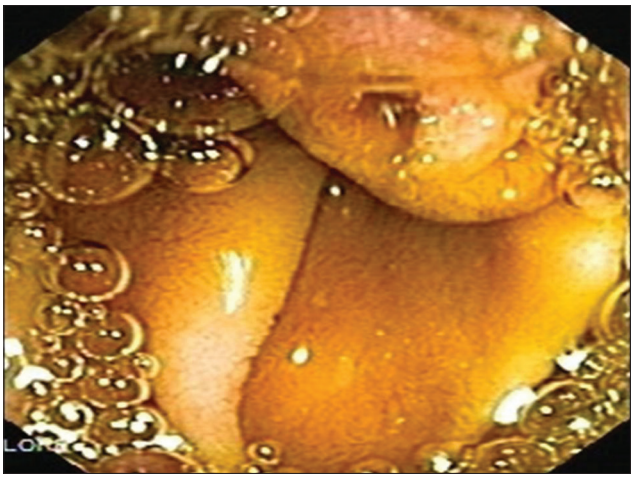

Figure 2: Endoscopic retrograde cholangiography-pancreatograph image of ampulla of vater

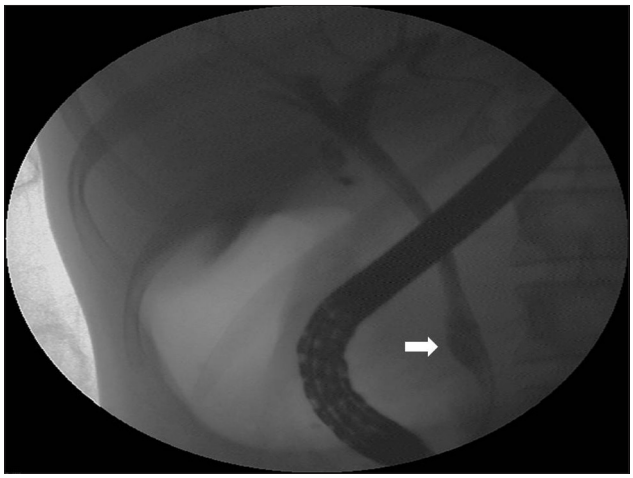

Figure 3: Endoscopic retrograde cholangiography showing dilatation of intramural portion of common bile duct

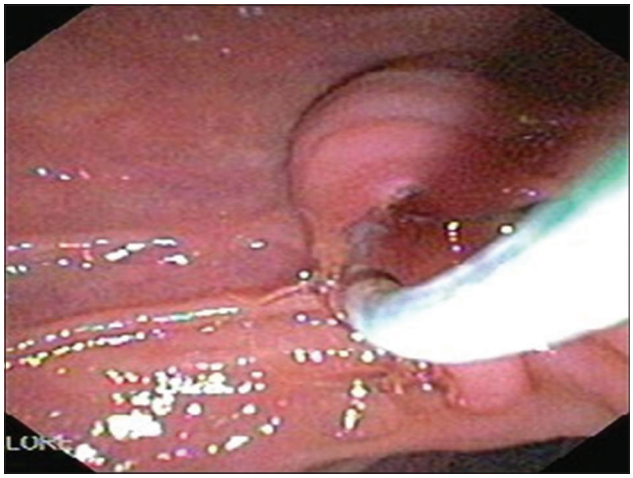

Figure 4: Endoscopic retrograde cholangiography-pancreatograph image showing biliary sphincterotomy

sphincterotomy, good drainage of bile was noticed. The patient gradually recovered well. At 24 months of follow-up, she is asymptomatic with no further episodes of pain.

Discussion

Acute pancreatitis according to revised Atlanta classification[1] can be defined as an acute inflammatory process of the pancreas, which requires two of three features like abdomen pain consistent with acute pancreatitis, more than three-fold rise in amylase and lipase, and imaging features of acute pancreatitis. Recurrent acute pancreatitis is characterized by two or more episodes of acute pancreatitis with complete or near complete improvement of clinical features of pancreatitis between episodes and without evidence of chronic pancreatitis.[2] The etiology of recurrent acute pancreatitis includes mechanical, metabolic, anatomical, and other miscellaneous causes. The various causes of anatomical abnormality comprise of pancreas divisum, anomalous pancreaticobiliary ductal union, annular pancreas, choledochal cyst, and duodenal duplication.[2] However apart from pancreas divisum, other causes are very rare.

The cystic dilation of the intraduodenal portion of CBD is known as choledochoceles. They are also classified as Type III choledochal cyst; however, they represent the least <5% of all choledochal cyst in asia.[3] In Jesudason *et al*. study of choledochal cyst of 15-year duration at Vellore, India, Type I was most common, and Type III was not reported.[4] The congenital choledochocele is either due to growth of rudimentary bile duct or could be due to congenital cyst or diverticulum of the intramural segment of the CBD. The acquired causes include papillary inflammation, stenosis, or secondary effect of sphincter of Oddi dysfunction leading to increased sphincter pressure and cystic dilatation.[5]

Takuma *et al*.[6] in a study of etiology of recurrent acute pancreatitis of 74 patients showed the presence of pancreaticobiliary malformation in 22% of patients. There

were three patients with choledochal cyst that comprises of 4% of patients of recurrent acute pancreatitis.[4]

On the basis of anatomic appearance, choledochoceles are classified into Type A and B by Sarris and Tsang.[7] Type A choledochoceles are characterized by cystically dilated intramural bile duct which open into the duodenum through a different opening. Type B choledochoceles are characterized as diverticulum of the intra-ampullary common channel, with bile duct opening normally into the duodenum.[7]

The choledochoceles commonly manifest clinically as multiple episodes of acute pancreatitis with complete resolution in between the episodes.[5,8] In comparison to other types of choledochal cyst, choledochoceles manifesting as jaundice and cholangitis are less frequent. The pathophysiology of choledochoceles causing acute pancreatitis includes: (1) Obstruction of pancreaticobiliary reflux resulting in increased pressure in duct or reflux of bile into the pancreatic duct. (2) Stone in cyst causing external compression of papilla resulting into pancreatitis. The incidence of malignancy in this type of choledochal cyst is lower than the other types. Our case also had a similar presentation of recurrent acute pancreatitis for last 2 years.[5]

The diagnosis of choledochoceles is based on cross-sectional imaging with computed tomography or magnetic resonance imaging. They reveal a distinct cystic structure on the medial wall of descending duodenum along with the size and anatomic relationship to the pancreaticobiliary tree.[5] EUS also helps in identifying wall layers of cyst as well as stones, sludge, or mucosal masses within it. It also helps in differentiating from duodenal duplication by finding continuity with duodenal wall layers.[5] Endoscopic retrograde cholangiopancreatography is widely used as a diagnostic tool, but it can also offer therapeutic potential during the same procedure. Endoscopic cholangiography shows retention of contrast medium into cystic dilation at the terminal end of CBD.[5] Type A choledochoceles can be treated using either endoscopic sphincterotomy or snare resection, while Type B should be completely removed by polypectomy snare.[5] Our patient was treated with biliary sphincterotomy, and she is doing well till 24 months with no episodes of abdominal pain.

Conclusion

Pancreaticobiliary malformations like choledochocele are rare anatomic cause of recurrent acute pancreatitis, which should be diagnosed early. Its early diagnosis will help to treat patients with endoscopic therapy. Hence, evaluation of pancreaticobiliary malformation should be an integral part of recurrent acute pancreatitis patients. Consideration should also be given to posttreatment surveillance of choledochoceles that are managed by endoscopic management, particularly in the young patient.[5]

References

1. Banks PA, Bollen TL, Dervenis C, Gooszen HG, Johnson CD, Sarr MG, *et al.* Classification of acute pancreatitis-2012: Revision of the Atlanta classification and definitions by international consensus. Gut 2013;62:102-11.
2. Kedia S, Dhingra R, Garg PK. Recurrent acute pancreatitis: An approach to diagnosis and management. Trop Gastroenterol 2013;34:123-35.
3. Edil BH, Cameron JL, Reddy S, Lum Y, Lipsett PA, Nathan H, *et al.* Choledochal cyst disease in children and adults: A 30-year single-institution experience. J Am Coll Surg 2008;206:1000-5.
4. Jesudason SR, Jesudason MR, Mukha RP, Vyas FL, Govil S, Muthusami JC. Management of adult choledochal cysts – A 15-year experience. HPB (Oxford) 2006;8:299-305.
5. Law R, Topazian M. Diagnosis and treatment of choledochoceles. Clin Gastroenterol Hepatol 2014;12:196-203.
6. Takuma K, Kamisawa T, Hara S, Tabata T, Kuruma S, Chiba K, *et al.* Etiology of recurrent acute pancreatitis, with special emphasis on pancreaticobiliary malformation. Adv Med Sci 2012;57:244-50.
7. Sarris GE, Tsang D. Choledochocele: Case report, literature review, and a proposed classification. Surgery 1989;105:408-14.
8. Ziegler KM, Pitt HA, Zyromski NJ, Chauhan A, Sherman S, Moffatt D, *et al.* Choledochoceles: Are they choledochal cysts? Ann Surg 2010;252:683-90.

How to cite this article: Patil M, Sheth KA. An uncommon evaluation in recurrent acute pancreatitis. J Dig Endosc 2014;5:116-8.

Source of Support: Nil, **Conflict of Interest:** None declared.