



Case Report

Duodenal Duplication Cyst with Recurrent Acute Pancreatitis: Report of a Case

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Abstract

Introduction: Duplication cyst can be occurred in any level of GI tract but the duodenal cysts are extremely rare. Most of the duplication cysts are detected in children. Duodenum duplication cysts are difficult to diagnose, as the presenting symptoms are nonspecific and are closely related to the type, size and location of the lesion. Both CT imaging and MRI can adequately identify a duodenal duplication cyst.

Presentation of the case: We report an adult with recurrent episodes of acute pancreatitis who was diagnosed with ERCP. With the diagnosis of duodenal duplication cyst, we planned to perform surgical resection of the cyst.

Conclusions: The surgical intervention for duodenal duplication cyst includes complete or partial surgical resection of the cyst. The location of the cysts in relation to the duodenum, especially to the ampulla, is important to determine the treatment strategy. Alternatively, duodenum duplication can be safely and effectively treated by different endoscopic interventions.

Keywords: Duplication cyst, pancreatitis, duodenal

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Introduction

Duplication cysts were first described by Fitz in 1884, and the nature of the disease was refined by Gross in 1953. Duplication cyst can be occurred in any level of GI tract but the duodenal cysts are extremely rare. Most of the duplication cysts are detected in children and fewer than 30% of all duplications are diagnosed in adults [1-3]. Duodenum duplication cysts (DDC) are difficult to diagnose, as the presenting symptoms are nonspecific and are closely related to the type, size and location of the lesion. And also DDC were reported to be responsible for duodenal obstruction, pancreatitis and gastrointestinal bleeding. Herein, we report a rare case of duodenal duplication with recurrent episodes of acute pancreatitis in an adult patient.



Figure 1 CT image of the cystic lesion

Case presentation

A 37-year-old female patient was admitted to the department of Gastroenterology in Ege University with the complaints of abdominal pain and nausea for three days. The patient suffered three episodes of acute pancreatitis in the last year and the etiology had not been identified by the usual diagnostic workup. In the last admission, physical examination showed a moderately distressed patient with a temperature of 36.6°C, a respiratory rate of 16 breaths/min, a blood pressure of 110/65 mmHg. Abdominal examination revealed diffuse tenderness without rigidity. Blood counts revealed signs of inflammation (white blood count (WBC): 14,600 g/L). Biochemical investigations showed increased serum levels of amylase (3349 U/L) and lipase (5308 U/L). She had no other significant laboratory results. An abdominal computed tomography (CT) revealed an enlarged and edematous pancreas with a dilated pancreatic duct (Fig. 1). And the distal part of the bile duct was compressed by a cystic lesion measuring 4.5x2.5x3.5 cm. MRCP revealed a cystic lesion measuring 4.5 cm that was located in the anterior part of the right kidney with the differential diagnosis of pancreatic pseudocyst or duodenal cyst. ERCP was performed and in the second duodenal portion a protruding duodenal mass that was compressing the ampulla Vateri was observed. With the diagnosis of duodenal duplication cyst, we planned to perform surgical resection of the cyst. A duodenotomy was performed, and the cystic lesion was observed under the mucosal layer in close proximity to the ampulla Vateri (Fig. 2). The duodenal mucosa adjacent

to the ampulla was incised longitudinally along the cystic lesion and the lesion carefully dissected from the submucosa. The common bile duct and main pancreatic duct were identified and cannulated with plastic catheters (Fig.3). After the excision the cystic lesion mucosal defect was closed. Sphincterotomy was performed to have a easier drainage of the common bile duct and the pancreatic duct. Duodenotomy was closed by primary sutures. Following an uneventful recovery, the patient was discharged 7 days later. The hystopathologic examination revealed a duodenal duplication cyst and there was no evidence of malignancy or dysplasia. After a 6 months follow up, she remains in good health.

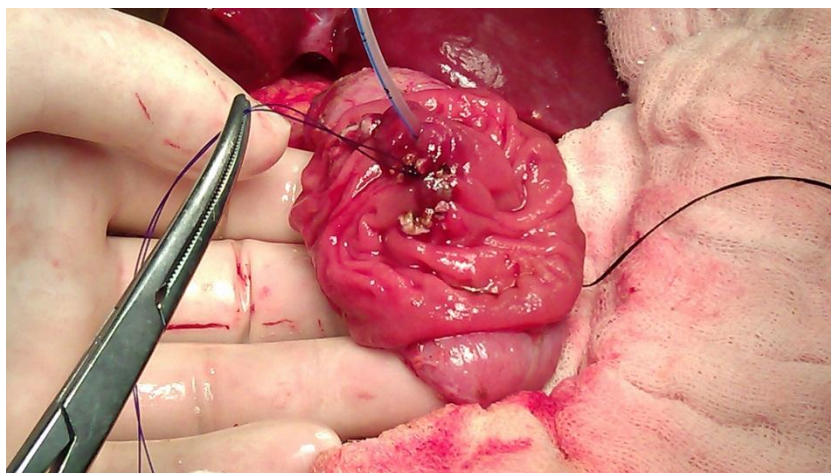


Figure 2 The cystic lesion under the mucosal layer in close to the ampulla vaterly

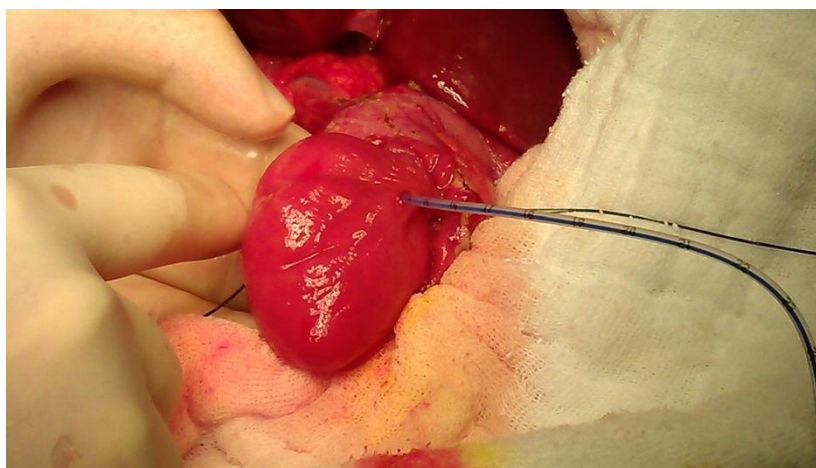


Figure 3 Cannulation of the common bile duct and main pancreatic duct

Discussion

DDCs are observed in less than 1/100,000 live births. Alimentary tract duplications are rare congenital anomalies that can occur anywhere along the alimentary tract from the mouth to the anus. The most

commonly affected sites are the small bowel (47%), colon (20%), esophagus (17%), stomach (8%), and duodenum (5%) [4]. Periapillary duplication cysts are extremely rare in duodenal duplication cyst cases, where discrimination from choledochocoele demands particular attention. The principle distinguishing features between periapillary DDC and choledochocoele are histopathological characteristics.

Clinical findings of duodenal duplication cysts may be non-specific, such as mild abdominal pain, or more specific, such as acute or chronic pancreatitis [5]. DDCs could stay silent for many years before they cause any symptoms including bowel obstruction, pain, distention or gastrointestinal bleeding. Bleeding, ulceration, and perforation may occur in the cysts containing ectopic gastric mucosa [6,7]. Many mechanisms could be responsible for pancreatitis: a transitory and mobility-related duodenal obstruction of the major papilla outflow by the cyst; the migration of biliary sludge and/or microstones from the cyst to the biliary tree, as observed in biliary pancreatitis; the communication of the dorsal pancreatic duct with the cystic cavity [5,8,9]. In the present case, the duodenal duplication cyst most probably caused mechanical obstruction of the pancreatic duct. Authors stated that both CT imaging and MRI can adequately identify a duodenal duplication cyst [10,11]. Also Antaki et al. stated diagnosis can be made without ERCP in most of the cases with ERCP and EUS [12]. However, due to the rarity of duodenal duplication cyst and lack of clinical experiences, more common lesions such as pancreatic pseudocyst are often suspected as the present case [13].

The surgical intervention for duodenal duplication cyst includes complete or partial surgical resection of the cyst. The location of the cysts in relation to the duodenum, especially to the ampulla, is important to determine the treatment strategy [10, 11,14]. Alternatively, duodenum duplication can be safely and effectively treated by different endoscopic interventions [10,12]. Duodenal duplication cysts are considered as benign clinical lesions, three cases with malign transformation were reported [2]. So a long-term follow-up is necessary for patients who had undergone partial resection of a duodenal duplication cyst.

In the present case, we described a very rare case of an adult with recurrent idiopathic pancreatitis. The diagnosis was periapillary duodenal duplication cyst that mimicking pancreatic pseudocyst. Both CT imaging and MRI can adequately identify the duodenal duplication cysts. Recurrent idiopathic pancreatitis directed to pancreatic pseudocyst and mislead us in CT imaging and MRI in the present case. The patient was diagnosed with ERCP that always not necessary for diagnosis.

Abbreviations

CT: Computed Tomography

MRI: Magnetic Resonance Imaging

EUS: Endoscopic Ultrasound

ERCP: Endoscopic Retrograde Cholangiopancreatography

MRCP: Magnetic Resonance Cholangiopancreatography

References

1. Browning RW. Duodenal duplications. *Rev Surg*. 1963, 20: 226-229
2. Chen JJ, Lee HC, Yeung CY, Chan WT, Jiang CB, Sheu JC. Meta-analysis: the clinical features of the duodenal duplication cyst. *J Pediatr Surg*. 2010, 45: 1598-1606

3. Ko SY, Ko SH, Ha S, Kim MS, Shin HM, Baeg MK. A case of a duodenal duplication cyst presenting as melena. *World J Gastroenterol*. 2013, 19:6490-6493
4. Macpherson RI. Gastrointestinal tract duplications: Clinical, pathologic, etiologic, and radiologic considerations. *Radiographics*. 1993, 13:1063-1080
5. Redondo-Cerezo E, Pleguezuelo-D áz J, de Hierro ML, Macias-S ánchez JF, Ubi ña CV, Mart ín-Rodr íguez Mdel M et al. Duodenal duplication cyst and pancreas divisum causing acute pancreatitis in an adult male. *World J Gastrointest Endosc*. 2010, 2:318-320
6. Gross RE, Holcomb GW Jr, Farber S. Duplications of the alimentary tract. *Pediatrics*. 1952, 9: 449-468
7. Rubin RB, Saltzman JR, Zawacki JK, Khan A, Swanson R. Duodenal duplication cyst with massive gastrointestinal bleeding. *J Clin Gastroenterol*. 1995, 21: 72-74
8. Hwang JH, Saunders MD, Rulyak SJ, Shaw S, Nietsch H, Kimmey MB. A prospective study comparing endoscopy and EUS in the evaluation of GI subepithelial masses. *Gastrointest Endosc*. 2005, 62:202-208
9. Carbognin G, Guarise A, Biasiutti C, Pagnotta N, Procacci C. Duodenal duplication cyst identified with MRCP. *Eur Radiol*. 2000, 10:1277-1279
10. Salemis NS, Liatsos C, Kolios M, Gourgiotis S: Recurrent acute pancreatitis secondary to a duodenal duplication cyst in an adult. A case report and literature review. *Can J Gastroenterol*. 2009, 23:749-752
11. Uzen MA, Koksall N, Kayahan M, Celik A, Kilicoglu G, Ozkara S: A rare case of duodenal duplication treated surgically. *World J Gastroenterol*. 2009, 15:882-884
12. Antaki F, Tringali A, Deprez P, Kwan V, Costamagna G, Le Moine O et al. A case series of symptomatic intraluminal duodenal duplication cysts: presentation, endoscopic therapy, and long-term outcome (with video). *Gastrointest Endosc*. 2008, 67:163-168
13. Yang M, Li DY, Zeng YM, Chen PY, Geng LL, Gong ST. Recurrent acute pancreatitis and massive hemorrhagic ascites secondary to a duodenal duplication in a child: a case report. *J Med Case Rep*. 2013 Mar 14;7(1):70. doi: 10.1186/1752-1947-7-70
14. Kawahara H, Takahashi T, Okada A: Characteristics of duodenal duplications causing pancreatitis in children and adolescents :a case report and review of the literature. *J Pediatr Gastroenterol Nutr*. 2002, 35:372-376