

## Case Report

# Isolated retroperitoneal enteric duplication cyst associated with an accessory pancreatic lobe

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**Abstract:** Introduction: Enteric duplication cysts are rare congenital anomalies. They are lined by gastrointestinal mucosa, connected to the digestive tract, and share smooth muscle layers and a common blood supply. In rare cases, duplication cysts are isolated from the digestive tract and have a unique blood supply. No patient with isolated duplication cysts that are located in the retroperitoneum and associated with an accessory pancreatic lobe at the onset have been reported to date. Materials and methods: A 10-year-old Asian boy complained of left upper abdominal pain for more than 3 months. Contrast-enhanced computed tomography showed that the main pancreatic duct in the tail of the pancreas was dilated. A soft tissue density shadow was observed around the tail of the pancreas. The lesion was connected to the main pancreatic duct and the blood was supplied from a branch of the splenic artery. Surgical exploration and pathologic specimens resulted in the diagnosis of an isolated retroperitoneal enteric duplication cyst associated with an accessory pancreatic lobe. The patient received treatments of rehydration, antibiotics, and protease inhibitors. Due to the poor conservative treatment effect in internal medicine, a surgical resection of abnormal tissue was performed. Results: The boy did not have abdominal pain again in the first year after leaving the hospital. Discussion: For repeated abdominal pain in young people, especially in children, an enteric duplication cyst needs to be ruled out. This case was difficult to diagnose and imaging examination was not able to determine whether it is located in the anterior peritoneum or the retroperitoneum. For such cases, surgical exploration is necessary, and surgical resection can achieve more satisfactory results.

**Keywords:** Retroperitoneal, isolated duplication, enteric duplication cyst, accessory pancreatic lobe

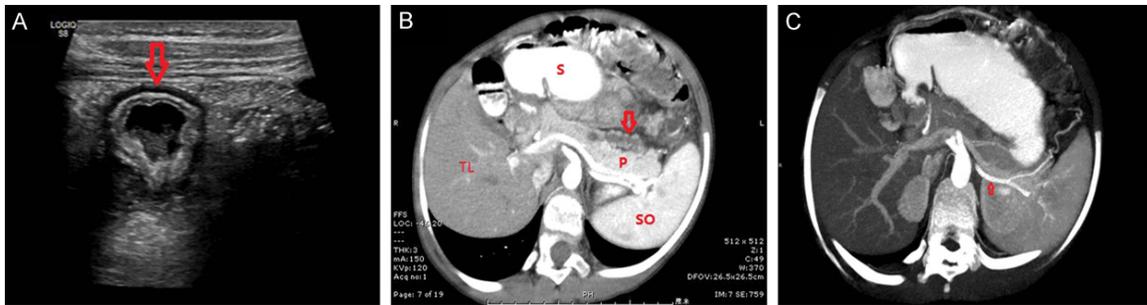
## Introduction

An enteric duplication cyst is an uncommon congenital abnormality that may occur anywhere between the mouth and rectum [1]. Enteric duplication cysts are usually anatomically connected to some portion of the gastrointestinal tract [2], but rare cases of isolated duplication cysts of the gastrointestinal tract have been reported. Among these cases, an associated accessory pancreatic lobe is an extremely rare congenital anomaly [3]. In patients with this anomaly, a definitive diagnosis is rarely made preoperatively, and several surgical explorations are necessary in some cases [4, 5]. Here, we report a case of an isolated retroperitoneal enteric duplication cyst associated with an accessory pancreatic lobe and describe the diagnosis, surgical treatment, and postop-

erative course of a patient with these anomalies. Furthermore, we review the literature regarding the clinical aspects of this anomaly. This work has been reported in accordance with SCARE criteria [6].

## Case presentation

A 10-year-old Asian boy was admitted to the hospital because of left upper abdominal pain for more than 3 months. In the previous 3 months, he experienced recurring upper left abdominal pain, which was described as paroxysmal cramps, accompanied by radiating pain in the left lower back. There was no abdominal distension, nausea, or vomiting. Physical examination showed left upper abdominal pain, blood biochemistry mainly showed increased blood amylase (1109 U/L, normal <600) and urinary amy-

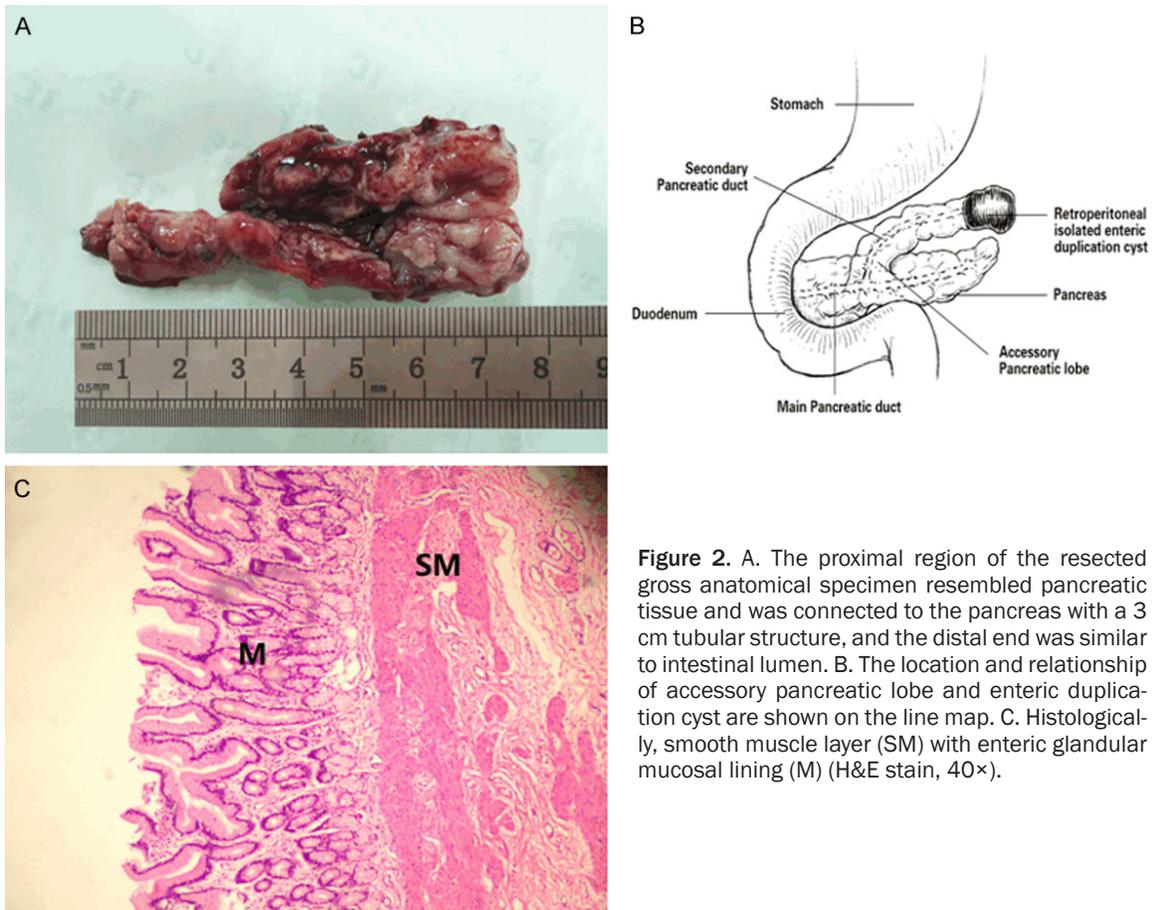


**Figure 1.** A. Color Doppler ultrasound view of the lesion: a cyst with the “double-wall” sign: the mucosa is hyperechoic (arrow) and the muscular layer is hypoechoic. An enteric duplication cyst was suspected. Abdominal enhanced CT showed that. B. The pancreatic duct was dilated in the accessory pancreatic lobe (arrow) and connected to the main pancreatic duct. C. The blood supply vessels of the deformed tissue are seen from the branches of the splenic artery (arrow), (TL: the liver, S: stomach, P: pancreas, SO: splenic organ).

lase (12589 U/L, normal <600), and blood glucose was normal; and blood glucose was normal. The boy had no history of trauma, specific medication use, or epidemiological exposure. The initial diagnosis was considered acute pancreatitis and treated by rehydration, antibiotics and protease inhibitors, after which the symptoms were slightly relieved. To further understand the cause of acute pancreatitis, abdominal color ultrasound (US) was performed and showed a thick-walled cystic structure with a size of approximately 27\*28\*23 mm in the abdominal cavity of the left upper abdomen. The distribution of annular blood flow signals could be seen on the wall; thus, intestinal repeat deformity was suspected (**Figure 1A**). Further contrast-enhanced computed tomography (CT) showed that the pancreas was not enlarged and that the main pancreatic duct in the tail of the pancreas was dilated. A soft tissue density shadow of approximately 31 mm\*25 mm was observed around the tail of the pancreas (**Figure 1B**). Enhancement CT showed that the lesion was connected to the main pancreatic duct and that the blood was supplied from a branch of the splenic artery (**Figure 1C**). We believed that communication between the lesion and the main pancreatic duct was the cause of pancreatitis. Because of the recurring left upper abdominal pain and poor effect of conservative treatment, we decided that surgery was the most appropriate choice for preventing the recurrence of acute pancreatitis.

In November 2017, the patient underwent laparotomy performed under general anesthesia. An approximately 10-cm incision was made through the rectus abdominis in the left upper

abdomen. In the left middle abdomen, a mass measuring approximately 7 cm\*3 cm in size with a nodular surface was found in the retroperitoneum extending to the posterior lateral side. We separated the mass, explored the bowel-like tissue, and observed that the tissue was connected to the tail of the pancreas. Rapid pathologic examination during surgery suggested that the tumor was a repeated malformation of the pancreas and intestinal tract, and no signs of malignancy were found. The tumor was considered a benign lesion; thus, surgery of the repeat terectomy of the pancreas and intestinal tract was performed. The blood supply to the lesion came from the splenic artery; therefore, the blood vessels were disconnected and ligated along with the tail of the pancreas, and their contact was hence disconnected. To prevent complications, such as pancreatic leakage, after surgery, the tail of the pancreas was again sutured, and the mass was simultaneously resected and completely removed. The distal end of the mass was a blind end that was not connected to the intestinal tract. The proximal region of the resected gross anatomic specimen resembled pancreatic tissue and was connected to the pancreas with a 3 cm tubular structure, and the distal end was similar to the intestinal lumen (**Figure 2A**). The location and relationship of the accessory pancreatic lobe and enteric duplication cyst are shown on the line map (**Figure 2B**). Final pathologic analysis confirmed the presence of an enteric duplication cyst associated with normal pancreatic tissue (**Figure 2C**). The patient was discharged 13 days postoperatively and followed up for a year after discharge. He was not



**Figure 2.** A. The proximal region of the resected gross anatomical specimen resembled pancreatic tissue and was connected to the pancreas with a 3 cm tubular structure, and the distal end was similar to intestinal lumen. B. The location and relationship of accessory pancreatic lobe and enteric duplication cyst are shown on the line map. C. Histologically, smooth muscle layer (SM) with enteric glandular mucosal lining (M) (H&E stain, 40×).

readmitted to the hospital for the same symptoms.

### Discussion

Enteric duplication cysts are rare congenital anomalies that are observed in approximately 1/100,000 births [7, 8]. These enteric duplication cysts can occur throughout the gastrointestinal tract and have a predilection for the jejunum and ileum (47-70%); however, they can also be found in the colon (20%), esophagus (17%), stomach (8%) and duodenum (2-12%) [9]. Enteric duplication cysts are diagnosed by the following accepted diagnostic criteria: the presence of a well-developed coat of smooth muscle; an epithelial lining representing some portion of the intestinal tract mucosa; and intimate anatomic association with some portion of the gastrointestinal tract [10, 11]. However, in rare cases, cysts can be isolated from the gastrointestinal tract and usually have a separate vascular pedicle. Menon et al. [12] suggested a cyst with no discernible communication or connection to the adjacent alimentary

tract but the presence of typical histopathologic features of a duplication cyst would qualify for a diagnosis of an isolated duplication cyst. These features enable a retroperitoneal cyst to be called an isolated retroperitoneal enteric duplication cyst. Approximately 10% of duplicate intestinal cysts are associated with congenital pancreatic diseases [13]. An accessory pancreatic lobe, a rare congenital disease of the pancreas, is pancreatic tissue with abnormal ducts produced by the pancreas. Accessory pancreatic lobes are often associated with enteric duplication cysts [14]. Our review of the literature revealed only 22 cases that have been reported to date since Bradbeer [15] described the first case in 1959 (Table 1). The specific causes of this anomaly are not yet clear, but we believe that the “neural bowel hypothesis” proposed by McLedtie [33] and Torma [34] is the best explanation for the formation of abnormal tissue in this patient. Intestinal mucosal metaplasia and in situ errors of cell differentiation [35] can lead to abnormal formation of ventral pancreatic buds during the fifth week of pancreatic development [20].

When the intestines rotate, pancreatic cells adhere to adjacent structures. Under the traction of the intestinal nerves, abnormalities can form at both ends of the axis, leading to the formation of an enteric duplication cyst and accessory pancreatic lobe.

We searched for papers written in English regarding this topic that have been published to date. The PubMed/MEDLINE, Google Scholar, Embase and Scopus databases were searched for the following key words: gastric duplication, duodenal duplication, enteric duplication cyst, foregut duplication, accessory pancreas and accessory pancreatic lobe. We found 22 case reports of enteric duplication cysts with accessory pancreatic lobes (**Table 1**). A total of 14 patients (63.6%) were female, and 8 patients (36.4%) were male, in accordance with 8 patients reported by Turkvatan et al. [23], among whom enteric duplication cysts predominated in females; however, the patient in our case was male. The first symptoms of most affected patients include abdominal, nausea and vomiting. In patients with enteric duplication cysts communicating with an aberrant pancreatic duct, abdominal pain is often related to acute pancreatitis. Acute pancreatitis caused by this anomaly has been reported in 16 cases (72.7%); 56.3% of these patients (9/16) suffered from recurrent attacks of acute pancreatitis. Oeda et al. [36] reported that there were no significant differences between patients with pancreatitis and those without pancreatitis in terms of either sex or age. The underlying cause of acute pancreatitis was hypothesized to be obstruction of the pancreatic duct by viscous mucus secretions, hemorrhage from ulceration, or biliary sludge [17, 19]. In our case, the main pancreatic duct located in the tail of the pancreas was dilated, and the main pancreatic duct was connected to the lesion. Acute pancreatitis may have been caused by blockage of the main pancreatic duct by hemorrhage from the ulcerated lesion. In addition, the type and location of enteric duplication cysts are also related to the patients' symptoms. In the case reported by Albert et al. [25], repeated vomiting may have been related to pyloric stenosis caused by a duodenal duplication cyst. In the case reported by Spence et al. [21], recurrent hematochezia may have been related to a connection between a gastric duplication cyst and the transverse colon. Of the 22 cases

retrieved, 21 (95.5%) were gastric duplication cysts, and 1 (4.5%) was a duodenal duplication cyst. However, our case is the first case of an isolated retroperitoneal enteric duplication cyst. Furthermore, the cyst was only connected to the accessory pancreatic lobe. Therefore, distinguishing between preoperative and pancreatic pseudocysts is very difficult.

Ultrasound (US) is the imaging modality of choice for the diagnosis of enteric duplication cysts. Typical US features of enteric duplication cysts are the double wall or muscular rim signs, which refer to the appearance of a cyst resembling the gastrointestinal tract with an echogenic inner rim, corresponding to the mucosa, surrounded by a hypoechoic rim, representing the smooth muscle layer [37, 38]. In our case, the duplication cyst showed the double wall sign: the mucosa was hyperechoic, and the muscular layer was hypoechoic. Computed tomography (CT) is also important in the diagnosis and treatment of enteric duplication cysts. CT can show the location, extent, complications, related abnormalities and anatomic relationship of the cysts to surrounding structures. In our case, CT showed that the cyst was connected to an accessory pancreatic lobe but not to the gastrointestinal tract. Magnetic resonance imaging is not a routine method for the diagnosis of enteric duplication cysts. Considering the patient's young age and difficulty cooperating for such an examination, no magnetic resonance imaging was performed. Surgery is a routine treatment for enteric duplication cysts. Surgical resection can not only relieve the symptoms but also prevent malignant transformation. The symptoms in this patient were obviously relieved after surgery, and the patient was not re-hospitalized for similar symptoms during 1 year of follow-up.

## Conclusion

An isolated retroperitoneal enteric duplication cyst is a unique type of enteric duplication cyst and an unusual phenomenon. Furthermore, this type of cyst associated with an accessory pancreatic lobe is even rarer. These abnormalities may be associated with the onset of pancreatitis and may cause diagnostic difficulty owing to their resemblance to pancreatic pseudocysts. Therefore, an early and accurate diagnosis of this anomaly is important because

## Isolated retroperitoneal enteric duplication cyst associated with an accessory pancreatic lobe

**Table 1.** Enteric duplication cyst with accessory pancreatic lobe as reported in the literature

First author [Ref], year	Age/Sex	Symptoms	Pancreatitis	Operation	Duplication cyst type
Bradbeer [15], 1959	17 y/M	Postprandial abdominal pain	Yes	Excision of duplication, external drainage of pancreas	Gastric duplication cyst
Brugsch [16], 1964	5 y/M	Abdominal pain	Recurrent	Resection of the cyst, resection of accessory pancreas	Gastric duplication cyst
Longmire [17], 1973	15 y/F	Abdominal pain	Yes	Pancreaticoduodenectomy	Gastric duplication cyst
Torma [18], 1974	14 d/M	Epigastric mass	Yes	Antrectomy, resection of aberrant pancreas	Gastric duplication cyst
Torma [18], 1974	7 m/F	Vomiting, poor weight gain, and abdominal mass	Yes	Excision of the cyst, resection of aberrant pancreas	Gastric duplication cyst
Traverso [19], 1975	32 y/F	Recurrent abdominal pain	Recurrent	Distal pancreatectomy	Gastric duplication cyst
Rosenlund [20], 1978	3.5 y/M	abdominal pain	No	Excision of the cyst, resection of aberrant pancreas	Gastric duplication cyst
Spence [21], 1986	8 m/F	Abdominal pain; intermittent, recurrent rectal bleeding	No	Cyst and tongue of pancreas excised; partial resection of the transverse colon	Gastric duplication cyst
Hoffman [22], 1987	19 y/F	Epigastralgia with nausea and vomiting	Recurrent	Excision of duplication and aberrant pancreatic lobe	Gastric duplication cyst
Lavine [23], 1989	6 y/F	Fever, epigastric abdominal pain, vomiting, and dehydration	Recurrent	Resection of the cyst, resection of aberrant pancreas	Gastric duplication cyst
Alessandrini [24], 1991	14 m/M	Massive rectal bleeding and vomiting	No	Simple excision	Gastric duplication cyst
Moss [4], 1996	9 y/F	Recurrent abdominal pain and hyperamylasemia	Recurrent	Excision of duplication, Roux-Y drainage of pancreas	Gastric duplication cyst
Whiddon [5], 1999	24 y/F	Bouts	Recurrent	Resection of duplication and aberrant pancreatic lobe	Gastric duplication cyst
Albert [25], 2000	8 y/M	Abdominal pain and vomiting	Recurrent	Excision of duplication and aberrant pancreatic lobe; partial resection of the omentum	Duodenum duplication cyst
Muraoka [26], 2002	46 y/F	Epigastralgia with nausea and vomiting	No	Excision of duplication (gastrectomy) and aberrant pancreatic lobe	Gastric duplication cyst
Shinde [27], 2009	49 y/F	Epigastric pain	No	Excision of duplication and aberrant pancreatic lobe	Gastric duplication cyst
Chin [28], 2011	11 d/F	Persistent emesis	No	Excision of duplication and aberrant pancreatic lobe	Gastric duplication cyst
Christians [29], 2013	43 y/M	Recurrent upper abdominal pain	Yes	Excise cyst; remove aberrant pancreas	Gastric duplication cyst
Aysel [30], 2014	29 y/F	Severe epigastric pain radiating to the back, nausea and vomiting	Recurrent	Excision of duplication and aberrant pancreatic lobe	Gastric duplication cyst
Abhilasha [31], 2015	5 y/F	Recurrent upper abdominal pain	Yes	Surgical excision of the gastric duplication cyst and accessory pancreatic lobe	Gastric duplication cyst
Shabtaie [14], 2017	6 y/M	Recurrent upper abdominal pain	Recurrent	Resection of the accessory pancreatic tissue and cystic structure	Gastric duplication cyst
Rousek [32], 2018	22 y/F	Abdominal pain	Yes	Partial gastrectomy, resection of aberrant pancreas	Gastric duplication cyst
Present case	10 y/M	Recurrent upper abdominal pain	Yes	Excision of duplication and aberrant pancreatic lobe	Retroperitoneal isolated enteric duplication cyst with an accessory pancreatic lobe

Note: y: year; m: month; d: day; M: male; F: female.

suitable surgical treatment may lead to a satisfactory outcome.

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### Disclosure of conflict of interest

None.

### Abbreviations

US, color ultrasound; CT, computed tomography.

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