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## ABOUT COVER

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The primary task of *WJCC* is to rapidly publish high-quality Autobiography, Case Report, Clinical Case Conference (Clinicopathological Conference), Clinical Management, Diagnostic Advances, Editorial, Field of Vision, Frontier, Medical Ethics, Original Articles, Clinical Practice, Meta-Analysis, Minireviews, Review, Therapeutics Advances, and Topic Highlight, in the fields of allergy, anesthesiology, cardiac medicine, clinical genetics, clinical neurology, critical care, dentistry, dermatology, emergency medicine, endocrinology, family medicine, gastroenterology and hepatology, geriatrics and gerontology, hematology, immunology, infectious diseases, internal medicine, obstetrics and gynecology, oncology, ophthalmology, orthopedics, otolaryngology, pathology, pediatrics, peripheral vascular disease, psychiatry, radiology, rehabilitation, respiratory medicine, rheumatology, surgery, toxicology, transplantation, and urology and nephrology.

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CASE REPORT

# Gastric duplication cyst communicating to accessory pancreatic lobe: A case report and review of the literature

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Author contributions: Rousek M and Ryska M performed a surgery and designed the report; Rousek M collected the patient's clinical data; Pintova J investigated a patient; Rousek M, Kachlik D, Nikov A, Pintova J and Ryska M wrote the paper.

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## Abstract

***BACKGROUND***

The combination of a gastric duplication cyst and duplicated part of the pancreas is an extremely rare developmental defect. The incidence in the population, or the clinical impact thereof, has not been uncovered. Symptoms are unspecific. Surgery is the treatment of choice. Timely diagnostics are of utmost importance, albeit they might be challenging at times. Being so rare, case reports are currently the only relevant source of information about the condition. Therefore each published finding is of a clinical impact.

***CASE SUMMARY***

Our work describes the case of a 22 year-old patient, who developed idiopathic acute pancreatitis. A computed tomography scan discovered liquid collection between the antrum of the stomach and the head of the pancreas. Initially, the collection was thought to be a pancreatic pseudocyst. Endoscopic ultrasound-guided transgastric drainage showed to have only a temporary therapeutic effect. Magnetic resonance cholangiopancreatography showed an accessory pancreatic lobe with a separate duct system. The accessory pancreatic lobe exited the body of the pancreas and

was in contact with the cystic collection. The patient was indicated for surgical resection. Within the surgery, an *en bloc* resection of the accessory pancreatic lobe was performed with the antrum of the stomach containing the gastric duplication cyst. No complications were observed in the surgery or thereafter. In the five months follow-up period, the patient was completely symptom free. Histopathological findings confirmed the gastric duplication cyst communicating to accessory pancreatic lobe.

***CONCLUSION***

This developmental defect is extremely rare. It can cause recurrent acute pancreatitis. Diagnostics are challenging. Surgery is treatment of choice.

**Key words:** Pancreatic resection; Accessory pancreatic lobe; Acute pancreatitis; Developmental defect; Gastric duplication cyst; Case report



**Core tip:** The combination of gastric duplication cyst and pancreatic duplications is an extremely rare developmental defect that can cause serious health problems in patients, including attacks of recurrent acute pancreatitis. Surgery is the treatment of choice, leading to complete resolution of symptoms in all published cases. Our paper summarizes the available information on this rare developmental defect, with a focus on potential for timely diagnostics and optimum treatment strategy. We also suggest unification of until now non-uniform anatomical nomenclature and we present a current knowledge of embryogenesis. We believe this information can help physicians in treatment of similar cases in future.

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## INTRODUCTION

Duplications of the digestive tract can be observed with an incidence of 1:4500 for the general population[1]. They can be found along the entire scope of the digestive tract. Their occurrence in the stomach is rare, and reported between 4% and 9%[2]. If gastric duplication cysts are symptomatic, symptoms are most commonly expressed through epigastric fullness, weight loss, anaemia, dysphagia, abdominal tenderness, or epigastric mass on physical examination[3]. Cases of adenocarcinoma arising from gastric duplication cysts have also been described[4]. The exact incidence of pancreatic duplication is unknown. It tends to be described following computed tomography (CT) scans in examinations of the abdominal cavity performed for diverse indications. In that case, no intervention is required[5]. However, cases of acute pancreatitis have been reported[6]. Duplication of the pancreas often exists in relation to developmental defects of other organs. The most common incidence described is in combination with a gastric duplication cyst[7]. The incidence, or the clinical impact of combined gastric and pancreatic developmental defect has not been uncovered. Relevant data about this anomaly is lacking. Case reports are the only relevant source of information. This is why each published case report has clinical importance. We searched for English written papers published thus far. PubMed/MEDLINE, Google Scholar, EMBASE and Scopus databases were searched for key words: "Gastric Duplication, Foregut Duplication, Accessory Pancreas, Accessory Pancreatic Lobe, Aberrant Pancreas, Bifid Pancreas, Fishtail Pancreas and Pancreatic Duplication". We found 24 case reports of contiguous gastric duplication cyst communicating to a duplicated part of the pancreas (Table 1).

## CASE PRESENTATION

The 22-year-old female Caucasian patient was admitted to the local hospital for a 2 d history of abdominal pain. The patient had no relevant medical history and there was no relevant family history. The patient worked as a bartender and used to smoke 10 cigarettes per day. The patient declared no allergies, was 170 cm tall and weighed 60 kg. Upon the admission a tenderness of the mesogastrium was observed. A Blood test showed leukocytosis 13.1 $\times$ $10^9$/L (ref. 4.0 $\times$ $10^9$/L-10.0 $\times$ $10^9$/L) and an elevation of pancreatic amylase level 246 U/L (ref. 13-53 U/L). The diagnosis of idiopathic, mild acute pancreatitis was established. The initial ultrasound screening had reported liquid collection near the head of the pancreas. Conservative treatment led to complete regression of symptoms, with laboratory test values returned normal. The patient was dismissed but recurrent episodes of abdominal pain, anorexia, and weight loss persisted. A CT scan was performed, which confirmed the presence of a peripancreatic, thick-walled collection (sized 5.6 cm $\times$ 2.8 cm $\times$ 2.7 cm), located between the head of the pancreas and the posterior wall of the antrum of the stomach (Figure 1). In the context of the recent pancreatitis, the collection was interpreted as a pancreatic pseudocyst. Given the persisting symptoms, a decision was made to perform an endoscopic ultrasound guided transgastric drainage of the pseudocyst. During gastrointestinal endoscopy a prominence of a posterior gastric wall was observed (Figure 2). Two plastic stents were introduced transmurally into the cavity of the cyst. The patient's symptoms receded completely. To clarify the relationship between the cyst and the pancreatic duct, magnetic resonance cholangiopancreatography (MRCP)

**Table 1 Case reports of gastric duplication cysts communicating to a duplicated part of the pancreas**

| Authors | Pancreatitis | Accessory pancreatic lobe | Symptoms to-surgery interval | Age | Sex | Final procedure |
|---|---|---|---|---|---|---|
| Brugsch[23], 1964 | Recurrent | Yes | 1 yr | 5 yr | M | Resection of the cyst, resection of accessory pancreas |
| Katz *et al*[24], 1967 | Recurrent | No | 9 mo | 34 yr | F | Partial resection of the cyst, vagotomy |
| Longmire *et al*[20], 1973 | Yes | Yes | 6 mo | 15 yr | F | Pancreaticoduodenectomy |
| Torma[25], 1974 | No | Yes | 1 d | 14 d | M | Antrectomy, resection of aberrant pancreas |
| Torma[25] 1974 | No | Yes | 2 mo | 7 mo | F | Excision of the cyst, resection of aberrant pancreas |
| Traverso *et al*[9], 1975 | Recurrent 13x | Yes | 9 yr | 32 yr | F | Distal pancreatectomy |
| Rosenlund *et al*[26], 1978 | No | Yes | 4 mo | 3.5 yr | M | Excision of the cyst, resection of aberrant pancreas |
| Black *et al*[27], 1986 | No | No | 1 d | 9 mo | M | Excision of the cyst, oversewn communicating duct |
| Spence *et al*[7], 1986 | No | Yes | 5 d | 8 mo | F | Excision of the cyst, resection of aberrant pancreas |
| Hoffman *et al*[28], 1987 | Recurrent 3x | Yes | 1 yr | 18 yr | F | Excision of the cyst, resection of aberrant pancreas |
| Lavine *et al*[29], 1989 | Recurrent 6x | Yes | 2 yr | 6 yr | F | Resection of the cyst, resection of aberrant pancreas |
| Bearzi *et al*[30], 1990 | Recurrent | No | 9 yr | 53 yr | F | Billroth I, distal pancreatectomy |
| Alessandrini *et al*[31], 1991 | No | Yes | 1 d | 14 mo | M | Excision of the cyst, resection of aberrant pancreas |
| Moss *et al*[32], 1996 | Recurrent | Yes | 7 yr | 9 yr | F | Excision of the cyst, Roux-en-Y limb drainage |
| Whiddon *et al*[33], 1999 | Recurrent | Yes | 4 yr | 24 yr | F | Excision of the cyst, resection of aberrant pancreas |
| Muraoka *et al*[22], 2002 | No | Yes | 10 yr | 46 yr | F | Distal gastrectomy, resection of aberrant pancreas |
| Hishiki *et al*[34], 2008 | No | No | 1 yr | 1 yr | M | Enucleation of cyst, oversewn communicating fistula |
| Shinde *et al*[35], 2009 | No | Yes | 4 mo | 49 yr | F | Resection of the cyst, resection of aberrant pancreas |
| Chin *et al*[36], 2011 | No | Yes | 1 d | 11 d | F | Enucleation of cyst, resection of aberrant pancreas |
| Oeda *et al*[21], 2010 | Recurrent 5x | No | 17 yr | 38 yr | F | Open cyst gastrostomy |
| Türkvatan *et al*[37], 2014 | Recurrent | Yes | 6 yr | 29 yr | F | Resection of the cyst, resection of aberrant pancreas |
| Christians *et al*[38], 2013 | Yes | Yes | 29 yr | 43 yr | M | Resection of the cyst, resection of aberrant pancreas |
| Jain *et al*[39], 2015 | Yes | Yes | 4 mo | 5 yr | F | Excision of the cyst, resection of aberrant pancreas |
| Shabtaie *et al*[40], 2018 | Recurrent 6x | Yes | 3 mo | 6 yr | M | Resection of the cyst, resection of aberrant pancreas |
| Our patient 2018 | Yes | Yes | 1 yr | 22 yr | F | Partial gastrectomy, resection of aberrant pancreas |

M: Male; F: Female.

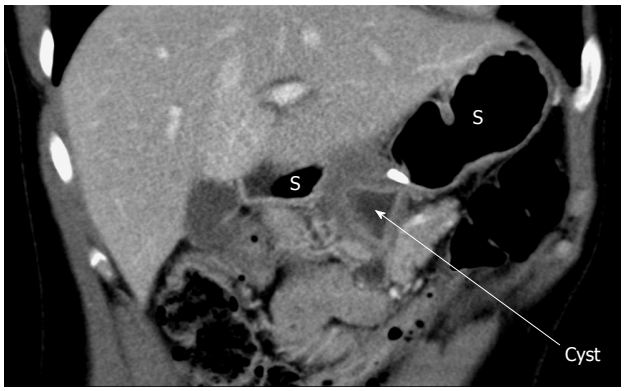


**Figure 1 Computed tomography scan of the abdominal cavity performed because of persisting abdominal pain.** Peripancreatic thick-walled collection (cyst) located between the head of the pancreas and the posterior wall of the antrum of the stomach (S). In the context of the recent pancreatitis, the collection was initially interpreted as a pancreatic pseudocyst. Published with permission of Luzicka nemocnice a poliklinika, Rumburk.

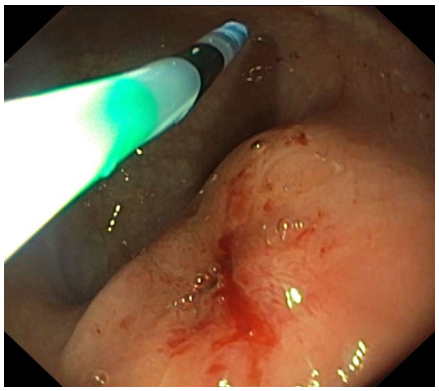

**Figure 2 The gastroscopy shows a prominence of a posterior gastric wall.** It represents a gastric duplication cyst located at antrum of the stomach. Published with permission of Jablonec nad Nisou Hospital.

was performed. Here, pancreatic duplication was described for the first time. The accessory pancreatic lobe was located ventrally and had a separate ductal system. The lobe communicated with the mass of the cyst localised between the pancreas and the antrum of the stomach. The pancreatic duct of the accessory lobe was connected to the main pancreatic duct (of Wirsung) within the body of the pancreas (Figure 3). The stents were extracted through an endoscopic procedure after 6 mo. The patient later again developed recurring attacks of abdominal pain and anorexia. Recurrence of acute pancreatitis was not observed.

## FINAL DIAGNOSIS

The patient was presented to our multidisciplinary team, which diagnosed a gastric duplication cyst communicating to an accessory pancreatic lobe.

## TREATMENT

Since all the case reports published thus far reported a complete resolution of symptoms after surgical treatment, it was indicated in this case as well (Figure 4). Given the existing, artificially created communication of

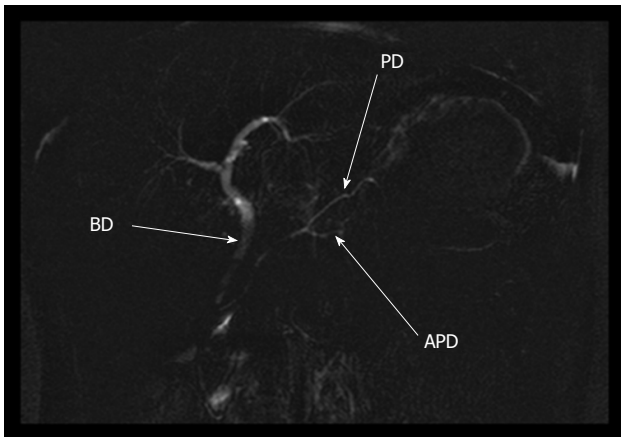


**Figure 3 Magnetic resonance cholangiopancreatography shows the pancreatic duplication.** The accessory pancreatic duct of the accessory pancreatic lobe is located ventrally to the main pancreatic duct. The confluence is located in the body of the pancreas. The biliary duct is of common appearance. Published with permission of Jablonec nad Nisou Hospital. APD: Accessory pancreatic duct; PD: Pancreatic duct; BD: Biliary duct.

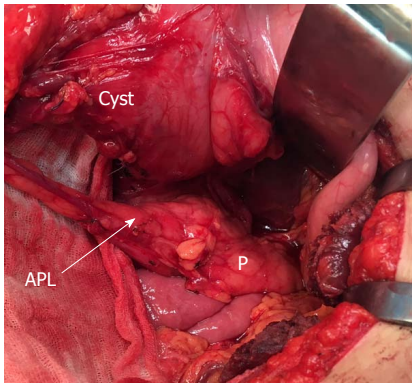


**Figure 4 Perioperative finding of the gastric duplication cyst communicating to accessory pancreatic lobe.** The accessory pancreatic lobe arising from the body of the pancreas. It follows a ventral direction to the stomach and communicates with a gastric duplication cyst (cyst) located at the antrum of the stomach. APL: Accessory pancreatic lobe; P: Pancreas.

the cyst cavity with the lumen of the stomach, it was technically impossible to separate the wall of the cyst from the antrum of the stomach during the surgery. We performed an *en bloc* antrectomy and stapler resection of the accessory pancreatic lobe in the spot where it was arising from the body of the pancreas.

## OUTCOME AND FOLLOW-UP

There were no complications within the surgery, and the patient was dismissed soon after. The patient showed no symptoms in the 5 mo follow-up period. The results of histopathological examination confirmed gastric duplication cyst with heterotopy of pancreatic tissue in its wall. The duplication communicated with an accessory pancreatic lobe of common histopathological appearance with a centrally located duct.

## DISCUSSION

As mentioned below a typical patient at time of diagnostics of gastric duplication cysts communicating to a duplicated part of the pancreas is a young female who suffered an episode of a mild-acute pancreatitis. Accompanying syndromes are usually abdominal pain, nausea and vomiting. The pancreatic duplication is mostly represented by an accessory pancreatic lobe and the gastric duplication cyst primary does not communicate to gastric lumen in most cases. Surgery leads to complete resolution of symptoms. Our patient met all these criteria. However we believe this article is unique, because to our knowledge, this is the first case report describing the attempt of endoscopic treatment, showing its only temporary effect.

The literature review indicated that all cases featured similar symptoms, challenging diagnosis, possible therapeutic solutions, and anatomical arrangement. The diagnostic criteria for gastric duplication cysts were set out in 1967. They comprise an inner digestive epithelial lining, an outer smooth muscle coat, blood supply originating from gastric vessels, and continuity with the gastric wall[8]. The fourth condition was eventually rescinded, and gastric duplication cysts can be divided, based on their relation to the gastric wall, either contiguous to the gastric wall or not contiguous to the gastric wall[9]. The gastric duplication typically originates from a wall of the stomach in the greater curvature area, or from the posterior wall of the stomach, and communicates with the pancreatic duplication. More often, in 80% of the cases (20/25), it communicates through an accessory pancreatic lobe. The lobe usually stems from the neck or body of the pancreas and follows a ventral direction. It crosses the pancreas and heads into the gastric duplication. In 20% (5/25) of cases, a simple bifurcation of the pancreatic ductal system in a macroscopically normal pancreas is present. In this case, the duplication cyst usually lies on the parenchyma of the pancreas, and the pancreatic duct of the accessory pancreatic lobe opens into it. Nomenclature has not been set in stone. Duplication cysts are better termed based on their anatomical location, rather than on their histopathological appearance. Therefore, *e.g.*, a gastric mucosa-containing cyst in the mediastinum is termed oesophageal duplication. Similarly, noncontiguous gastric cysts located in the pancreas are called pancreatic duplication cysts or enteric duplication cysts of the pancreas[10]. Other authors have used the term "foregut duplication cyst of the stomach" to describe a gastric duplication cyst[11]. The following terms can be found in the literature to denote pancreatic duplications: bifid pancreas, accessory pancreas, accessory pancreatic lobe, fishtail pancreas, bifid tail of pancreas, aberrant pancreas, and aberrant pancreatic lobe. We see "gastric duplication cyst communicating to accessory pancreatic lobe" as the most suitable term for this combined

developmental defect. Although not really a lobe in the true sense of the word, we believe this descriptive name best reflects the anatomical arrangement. In case the accessory pancreatic tissue is missing, we propose "gastric duplication cyst communicating to bifurcated pancreatic duct".

How this developmental defect arises is not fully understood. There are numerous theories, but the overall most accepted explanation for the development of the duplication of the digestive tract is split notochord syndrome. During week 3, the notochord and endoderm fuse and form the notochordal plates. Then, the mesoderm forms a continuous layer separated from the notochord. If the separation is impaired, an ecto-endodermal adhesion (neuroenteric band) can develop and persist. As it does not elongate as quickly as the adjacent structures, it causes traction diverticular structures of the intestine followed by the formation of duplication cysts located dorsally[12,13]. In week 5 of development, two pancreatic buds (ventral and dorsal) outpouch from the distal end of the endodermal foregut. The ventral pancreatic bud appears together with the primordium for the liver and biliary ducts (hepatopancreatic bud) and the dorsal pancreatic bud gives rise to the majority of the gland[14]. Later, both merge in the region of the pancreatic neck and form the final pancreas in week 7[15]. The dorsal pancreatic duct forms the pancreatic duct of the tail, body, neck, and, except for the terminal part of the pancreatic duct of Wirsung, also the head of the pancreas. The terminal part of the former dorsal pancreatic duct usually remains as a separate accessory pancreatic duct (of Santorini), patent in less than half of the population (43%)[16]. At the beginning, the ventral pancreas has two lobes with two primitive ducts[16]. Only the right lobe finally develops in the dorsocaudal part of the head of the pancreas and its primitive duct in the terminal part of the pancreatic duct of Wirsung. The other lobe and its duct regress and produce the uncinate process of the pancreas. This bilobular stage of the ventral pancreas as well as different proportional growth of the duct of the ventral pancreas, bile duct, and duodenal wall are considered as the crucial points for potential developmental disorders concerning the ductal defects, which development of has unfortunately not been thoroughly studied[5]. The ductal disorders appear later during development, after fusion of the ventral and dorsal pancreases when the primitive system of ducts develops. Concerning the accessory pancreatic tissue, four variants are relevant: the bifurcated pancreatic duct in the opposite direction, the trifurcated, the quadrifurcated, and the fish tail variant[17,18]. In the case of gastric cyst communicating with the ductal system of the pancreas, the dorsal traction on the distal portion of the foregut from which the dorsal pancreatic bud outpouches seems to be the trigger for cyst formation[19]. This developmental defect is most commonly diagnosed in young patients, with a median of 9 years (minimum of 11 days and maximum of 53 years). More than two thirds of the patients, 68% (17/25), were women. Diagnostics are challenging. This is due to the extremely rare incidence in the population and the non-specific symptoms. Recurrent acute pancreatitis forms the only partly pathognomonic symptom, as 60% (15/25) of patients suffered at least one episode of acute pancreatitis, 80% out of these (12/15) suffered recurrent attacks of acute pancreatitis. The literature describes a case of 13 episodes of acute pancreatitis in a single patient before proper diagnosis[9]. The cause of acute pancreatitis in these patients has not been fully explained. Obstruction of the pancreatic duct by blood, with subsequent haemoductal pancreatitis, is the prime suspect[20]. Alternatively, it may be caused by the obstruction of the pancreatic duct by mucoid secretion from the gastric duplication cyst with subsequent stasis of the pancreatic juice[9]. Therefore, communication between the duct of the accessory pancreatic lobe with the duplication cyst seems to be the predisposing factor. We must admit that acute pancreatitis can occur in patients where communication between the pancreatic duct and the duplication cyst has not been confirmed. The question remains whether, in these particular cases, it is impossible to prove an existing communication, or whether pancreatitis occurs through a different mechanism, for instance by ulceration passing through the cyst wall into the pancreatic parenchyma, which is caused by acid secretion of gastric glands that line the wall of the duplication[21]. It is worth noting that none of the patients developed severe, necrotising pancreatitis. All cases involved mild, self-limited diseases with a quick tendency for improvement of the general condition given conservative therapy. Other described symptoms include recurrent abdominal pain, vomiting, nausea, weight loss, failure to thrive, anorexia, palpable abdominal mass, and bleeding into the digestive tube. The foregoing indicates that young patients suffering an idiopathic acute pancreatitis should be thoroughly examined, including a MRCP screening, to rule out developmental defects of the pancreatic ductal system.

Initially, our case was also being deemed a post-pancreatic pseudocyst on the basis of the ultrasound and CT scans of the duplication cyst of the stomach. The diagnosis of an accessory pancreatic lobe was set on the basis of a review MRCP examination after a successfully endoscopic ultrasound-guided transgastric drainage of the cystic collection after which the patient reported a complete loss of symptoms. The abdominal pain became recurrent after the drain was extracted. This supports the opinion that the creation of gastropseudocystostomy may cause the patient's symptoms to disappear[21], similarly to the evacuation of the gastric duplication cyst content into the lumen of the stomach[22].

Nevertheless, surgery is recommended as the preferred treatment. First, it has led to the complete loss of symptoms in all published cases, including

cases of carcinoma arising from the cells of gastric duplication[4]. The published case reports most often entailed a simple resection of the duplication cyst, along with the resection of the accessory pancreatic tissue. In one case, a Roux-Y pancreaticojejunal anastomosis supplemented the resection. Left-sided pancreatectomy was performed in two cases, and pancreaticoduodenectomy was performed in one case. Resection of the distal part of the stomach was performed in four patients, gastrocystostomy was created in one case. In the majority of cases, gastric duplication cysts did not communicate with the lumen of the stomach. In fact, in our data set a communication was proven only in 8% (2/25) of all cases.

## CONCLUSION

Gastric duplication cyst communicating to a duplicated part of the pancreas is an extremely rare developmental defect. Relevant literature is lacking due to low prevalence. Timely diagnostics are challenging. Symptoms are unspecific, but most patients suffered acute pancreatitis. Therefore young patients suffering an idiopathic acute pancreatitis should undergo MRCP screening to rule out developmental defects of the pancreas. Endoscopic ultrasound-guided transgastric drainage seems to have only temporary therapeutic effect. Surgery is the preferred treatment. A simple resection of the gastric duplication cyst, without the need to enter the lumen of the stomach, seems to be the method of choice. This should be supplemented by a stapler resection of the accessory pancreatic lobe (if any) or by oversewing of the entry of the bifurcated pancreatic duct.

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