

## CASE REPORT

# A SEVERE CASE OF ACUTE NECROTIC PANCREATITIS CAUSED BY DUODENAL CYST DUPLICATION

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### ABSTRACT

**Introduction.** Duodenal duplication cyst (DDC) is a rare congenital malformation that appears in the embryonic development of the digestive tract. It is a benign condition usually diagnosed in infancy and early childhood, being a rare and difficult diagnosis in adult population. DDC is a recognized cause of duodenal obstruction, acute pancreatitis, obstructive jaundice and even digestive hemorrhage.

**Case presentation.** We report the case of a young adult male with abdominal pain history, who presents with recurrent episodes of acute severe necrotic pancreatitis. The abdominal computed tomography scan revealed a Balthazar C necrotic pancreatitis with partial thrombosis of the splenic vein and a cystic mass in the second part of the duodenum. The endoscopic ultrasonography (EUS) established that the duodenal cystic lesion came from the second layer, meaning the submucosa. We performed endoscopic cystotomy with complete evacuation of the fluid content into the duodenum, with favorable clinical outcome.

**Conclusions.** The particularity of the case is represented by the low incidence of this pathology and the rare form of presentation, meaning acute pancreatitis probably from pancreatic ductular hypertension caused by the DDC.

### RÉSUMÉ

**Un cas grave de pancréatite nécrotique aiguë causée par une duplication des cystes duodénaux**

**Introduction.** La duplication kystique duodénale est une malformation congénitale qui survient au cours du développement embryonnaire du tube digestif. C'est une pathologie bénigne diagnostiquée le plus souvent en néonatalogie ou dans l'enfance, et le diagnostic chez l'adulte est difficile et non spécifique. La DDC est une cause reconnue d'obstruction digestive, de pancréatite aiguë, d'ictère obstructif et, rarement, d'hémorragie digestive haute.

**Présentation de cas.** Nous rapportons le cas d'un adulte de sexe masculin aux antécédents de douleurs abdominales et d'épisodes récurrents de pancréatite aiguë. La CT de balayage a montré une pancréatite aiguë sévère Balthazar C avec une thrombose partielle de la veine splénique et une masse kystique au niveau de la seconde partie du duodénum. La formation sous-épithéliale écho-endoscopique était la sous-muqueuse II. On a réalisé une cystotomie endoscopique avec l'évacuation complète du liquide aux résultats cliniques favorables.

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**Key words:** duodenal cyst duplication, severe acute necrotic pancreatitis, endoscopic cystotomy.

**Conclusions.** La particularité du cas est représentée par la faible incidence de la pathologie et la forme rare de la présentation, c'est-à-dire une pancréatite aiguë due à l'hypertension canalaire pancréatique provoquée par la DDC.

**Mots-clés:** duplication kystique duodénale, pancréatite nécrotique aiguë sévère, cystotomie endoscopique.

## INTRODUCTION

Duodenal duplication cyst (DDC) is a rare congenital malformation that appears in the embryonic development of the digestive tract. It is a benign condition usually diagnosed in infancy and early childhood, being a rare and difficult diagnosis in adult population, because of the nonspecific symptoms related to the size and location of the lesion<sup>1</sup>. Duodenal cysts are very rare, the most common site of gastrointestinal duplications is in the ileum, the esophagus, and the colon. Duplication cysts are surrounded

with a gastrointestinal epithelial lining<sup>2</sup>. They are commonly tubular or cystic and they may communicate with the pancreatic and biliary system; in rare cases, the communication with the digestive lumen is present<sup>3</sup>. DDC is a recognized cause of duodenal obstruction, acute pancreatitis, obstructive jaundice and even digestive hemorrhage. The treatment of symptomatic DDC was traditionally surgical but now, due to the new endoscopic techniques, this can be done without considerable morbidity<sup>4</sup>.

## CASE PRESENTATION

A 43-year-old male patient presented at our hospital with intense epigastric and back pain, associated with nausea and vomiting. His medical history was positive for recent recurrent necrotic pancreatitis. At clinical examination, a raised heart rate, cold sweat hands and diffuse abdominal tenderness were observed. The blood tests revealed inflammatory syndrome with leukocytosis (14 630/mm<sup>3</sup>), hyperfibrinogenemia and pancreatic reaction, with elevated amylase and lipase (1132 U/L, 2112 U/L, respectively); renal and hepatic function tests were within normal ranges.

The abdominal ultrasonography showed a fat liver, with normal gallbladder and biliary tree, and



**Figure 1.** Abdominal ultrasound image showing pancreatic pseudocyst.



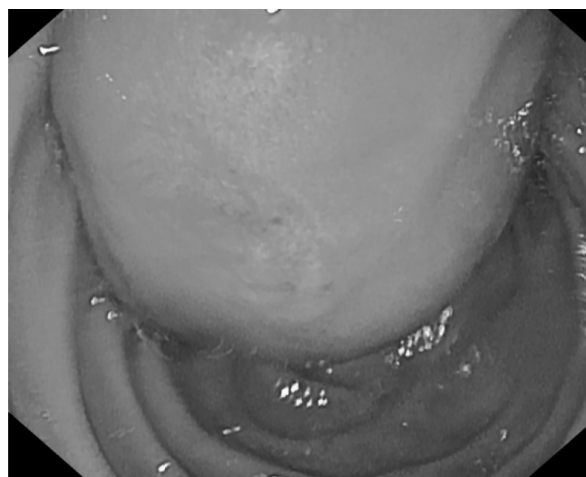
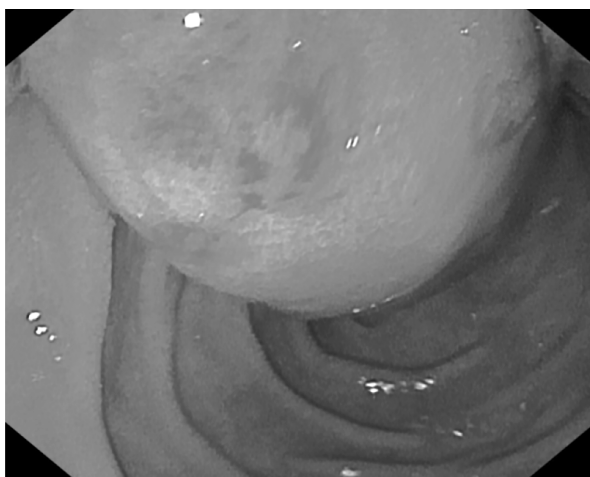
**Figure 2.** EUS image of the pancreas



**Figure 3.** EUS image of the pancreatic pseudocyst



**Figure 4 and 5.** Endoscopic ultrasonography demonstrating the cystic duodenal lesion surrounded by the digestive layers.



**Figure 6 and 7.** Upper gastrointestinal endoscopy demonstrating an extrinsic mass lesion with an intact mucosa protruding into the lumen of the second part of the duodenum.

a fluid cystic collection in pancreatic area (Figure 1). The abdominal computed tomography scan revealed a Balthazar C necrotic pancreatitis with partial thrombosis of the splenic vein, a cystic mass measuring 50mm x 40mm in size in the second part of the duodenum, and a fluid necrotic collection, measuring 90 mm in diameter (a „walled off pancreatic necrosis“).

We performed an upper digestive endoscopy, that revealed a huge extrinsic well-defined mass, with intact duodenal mucosa, bulging in the second portion of the duodenum (Figure 2 and 3).

The next step was to perform an endoscopic ultrasonography (EUS) to establish the adherence of the lesion. Linear EUS confirmed the presence of an enlarged pancreas, with pancreatitis criteria (Figure 4), a pancreatic pseudocyst (Figure 5) and an anechoic homogenous oval lesion, arising from the submucosal

layer of the duodenum wall, meaning a duodenal duplication cyst (Figure 6 and 7). We decided to perform the evacuation of the DDC with a cystotome, under echo-endoscopic guidance; at the end of the procedure, complete evacuation of the fluid content into the duodenum was obtained (Figures 8, 9 and 10). The clinical outcome of the patient was favorable in time, without recurrent pancreatitis.

## DISCUSSION

Digestive duplication cysts are rare congenital anomalies that can occur at any site of the digestive tract, most frequently in the ileum and rarely in the duodenum. There are three conditions necessary for the diagnosis of a duplication cyst: the lesion has to be adjacent to the gastrointestinal wall; it must be

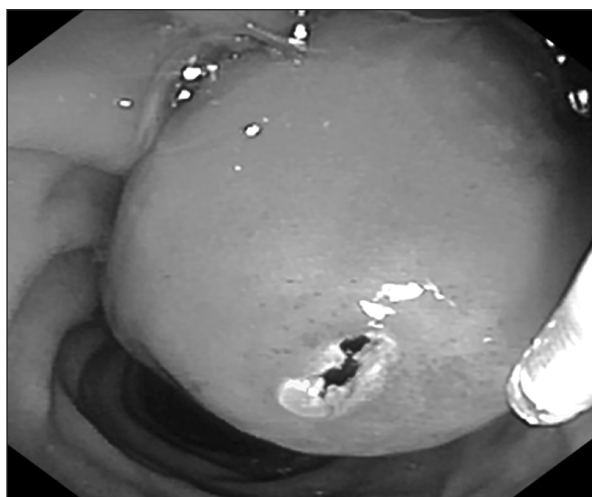


Figure 8 and 9. Endoscopic incision of the DDC with a cystostome, evacuating the cavity in the duodenal lumen.



Figure 10. Postprocedural image.

surrounded by a smooth muscular layer, and lined by a mucosal alimentary membrane<sup>5</sup>.

The diagnosis is frequent in children, but DDC can remain clinically silent until adulthood, when it becomes clinically obvious through complications. DDC can cause abdominal pain, vomiting, gastric or small bowel outlet obstruction, digestive hemorrhage, jaundice or pancreatitis<sup>6,7</sup>. The dimensions of the cyst vary from 2 cm to 5 cm in diameter.

The differential diagnosis of DDC is made with other cystic lesions in this area, such as pancreatic pseudocysts, cystic tumor of the pancreas, duodenal diverticula, cyst of the biliary tree, meaning choledochocoele or mesenteric cysts<sup>8</sup>. The layered pattern of the wall observed by the EUS makes the difference between the other forms of cystic lesions<sup>2,8</sup>.

The diagnosis is based on findings at contrasted enhanced tomography and endoscopic ultrasound identifying the type, location and size of the cyst<sup>9-11</sup>.

For biliary communication, magnetic resonance imaging (RMN) or endoscopic retrograde cholangiopancreatography (ERCP) examinations are useful<sup>4,8</sup>.

Total excision of the DDC is the preferred treatment. Surgical techniques were abandoned because of the raised morbidity, surgical complications and the possibility of damaging the biliary and pancreatic duct<sup>3</sup>. Endoscopic treatment is a safer and effective alternative to the surgical intervention<sup>5,12</sup>.

Malignant transformation of the residual tissue is a very rare complication reported in a few cases. The patient with partial removal or endoscopic treatment of a DDC needs long term follow-up<sup>13</sup>.

## CONCLUSIONS

The particularity of the case is represented by the low incidence of the pathology and the rare form of presentation, meaning acute pancreatitis likely from pancreatic ductular hypertension caused by the DDC.

Duodenal duplication should be taken into consideration when a cystic lesion near the digestive tract causes vague upper abdominal symptoms or other above mentioned complication. The treatment is always total excision, preferably using an endoscopic technique. The outcomes of these lesions are always favorable when treated radically.

## Compliance with Ethics Requirements:

„The authors declare no conflict of interest regarding this article“

„The authors declare that all the procedures and experiments of this study respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well as the national law. Informed consent was obtained from the patient included in the study“

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