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Case Report,

Intrapancreatic Peristaltic Duodenal Duplication Cyst; Presenting with Acute Pancreatitis and Intraabdominal Bleeding

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Abstracts:
Congenital anomalies and variations should be kept in mind when evaluating diseases in the pediatric patient group. A 12 years old girl presented with findings of acut abdomen. Clinical and laboratory data were consistent with acute pancreatitis. Imaging findings showed a peristaltic cystic tissue with a thick wall and central lumen in the head of the pancreas. In the operation, it was shown that this cystic lesion was associated with the accessory pancreatic duct and had no duodenal connection. In the pathology report, the presence of duodenal intestinal tissue compatible with a duplication cyst located in the head of the pancreas was proven. By sharing this rare case, keeping in mind the underlying congenital anomalies in any pathology in children and emphasizing the importance of ultrasonographic dynamic real-time examination in abdominal imaging.

Keywords: duplication cyst, intrapancreatic, pancreatitis, pediatric radiology, peristaltic cyst, ultrasonography

Brief Key Points: When evaluating pediatric patients, congenital anomalies should be kept in mind. A good ultrasound evaluation makes a great contribution to the diagnosis in children especially with low intra-adominal fat.

Introduction:

The etiology of acute pancreatitis in children is often trauma, infections, drugs, genetic factors and metabolic diseases. Rarely, congenital anomalies of the pancreaticobiliary duct system may cause pancreatitis such as pancreatic divisum and annular pancreas. Intrapancreatic enteric duplication cysts are extremely rare that may mimic pancreatic pseudocysts with recurrent pancreatitis [1].

Enteric duplication cysts are rare congenital malformations most commonly seen in the small intestine, especially in the ileum. They can occur in any part of the gastrointestinal (GI) system. The cysts are lined by GI epithelium, surrounded by smooth muscle walls. Most duplication cysts are detected in childhood and less than 30% are diagnosed in adulthood [2].

The diagnosis of enteric duplication cysts is difficult due to their variable clinical presentation. Usually, patients are asymptomatic and these lesions are often detected incidentally. Less frequently, patients may present with a complication such as intestinal

obstruction, intussusception, volvulus, pancreatitis, perforation, gastrointestinal bleeding, or infarction.
Enteric duplications are rarely occur in the head of the pancreas and presents with pancreatitis. We present a case of a child with pancreatitis and intraperitoneal bleeding because of intrapancreatic duodenal duplication cyst in the head of pancreas.

Case Report:

A 12 years old girl presented with abdominal swelling, epigastric pain and vomiting. There was no history of abdominal trauma, surgery and metabolic abnormality. She had intermittent abdominal pains for 5-6 years and a loss of 13 kg in the last 5 months.
Abnormal findings in the initial laboratory study were: Wbc: 15.400 /mm3 (4000-10000), Hgb: 4.9(11-15), Amylase 1334 U/lt (25-125), Lipase 697 U/lt (8-78), CRP: 6.5 mg/l (0-5), Sedimentation 26 mm/hour (1-20).

Radiologic Findings:

Ultrasonographic examination revealed a thick-walled cystic lesion in the central part of the head of the pancreas. During the examination, it was noticed that this lesion made peristaltic movements like intestinal organs (Figure 1). In the anterior neighborhood of this cystic mass, there was another cystic lesion with 16x10 mm dimensions, compatible with the pseudocyst, which did not have a prominent wall structure. A thin tract was observed between these two defined lesions (Figure 2).

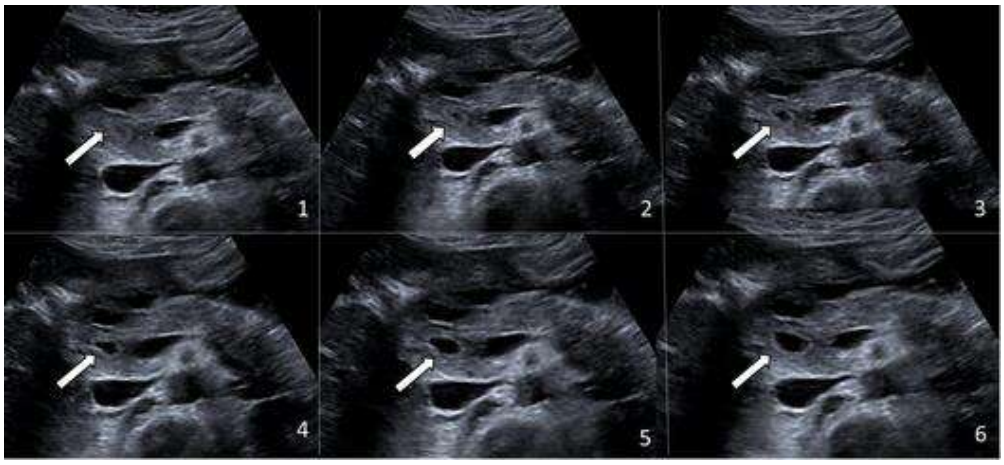

Figure 1: In the abdominal US images, the peristaltic movement of the thick-walled cystic lesion in the central pancreatic head is shown in the images given sequentially (1-6).

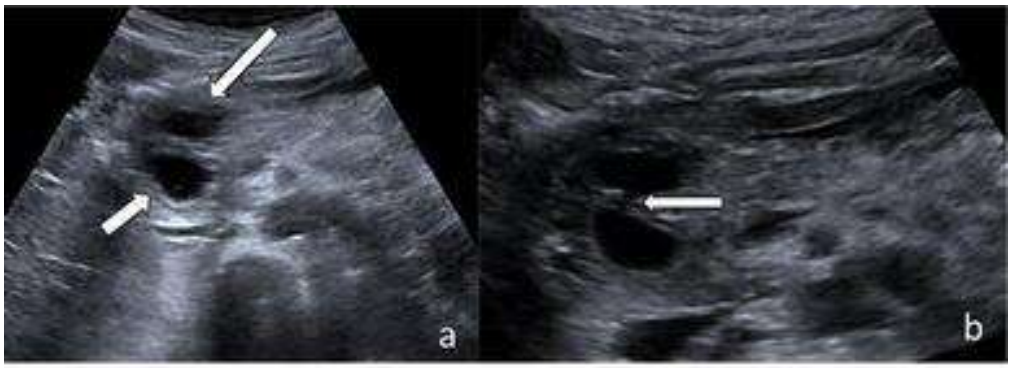

Figure 2: Abdominal US images shows a pseudocyst (long arrow) ,that is located anterior to the peristaltic duplication cyst (short arrow) (a). Also tehere is a thin tractus (arrow) between the peristaltic lesion and pseudocyst (b).

A difference in the size and appearance of the thick-walled cystic mass located in the pancreatic head was detected in successive T2-weighted axial MR images taken with one-minute intervals. When evaluated together with ultrasonographic data, it was understood that this was secondary to peristalsis (Figure 3). There was also diffuse ascites in all quadrants of the abdomen and minimal pleural effusion in both hemithorax.

Endoscopic examination revealed hemorrhagic material from the ampulla of Vater. Fluid as serohemorrhagic exudate was aspirated from the abdomen. Disseminated intravascular coagulation (DIC) and multiple organ dysfunction developed during clinical follow-up. When the clinical situation stabilized, it was decided to operate with the decision of the council. At surgery a cystic lesion with a diameter of approximately 15 mm, whose wall structure was selected in the central part of the pancreatic head and hemorrhage from this area to the abdomen were detected. During the operation, the accessory pancreatic duct associated with this cavitary lesion was visualized under fluoroscopy. There was no comminication with the main pancreatic duct and duodenum. Afterwards, the cystic mass was excised and drain catheters were placed, and the operation was terminated (Figure 4a, 4b).

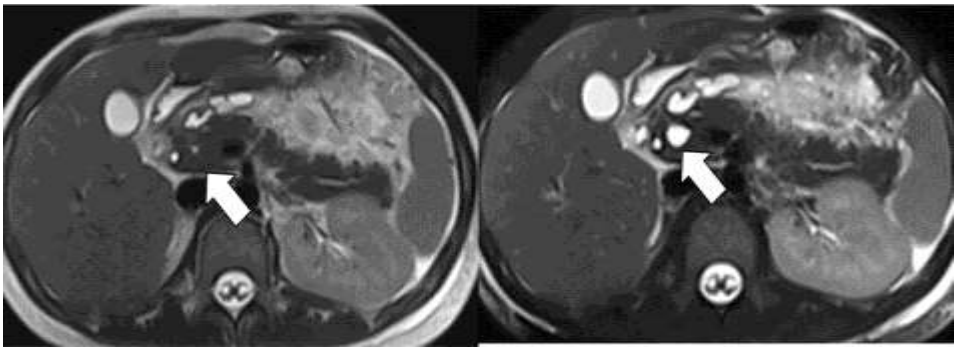

Figure 3: Sequential T2W images show mobile luminal tissue compatible with duplication cyst (short arrow), pseudocyst anteriorly (long arrow) and distal end of common bile duct (arrowhead) medially. In the examination performed with 1 minute intervals, changes in the wall thickness and luminal aperture secondary to peristalsis are noticed in the lesion in the central pancreatic head.

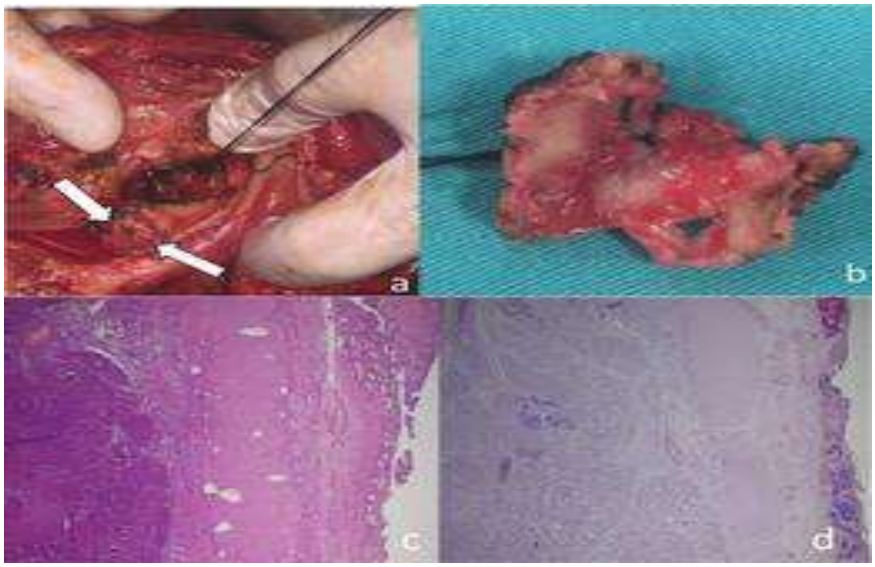

Figure 4: In the photographs taken during the operation, the tissue with a thick wall and lumen in the head of the pancreas was visualized(a). The excised duplication cyst with some pancreatic tissue is shown(b).In the microscopic evaluation, pancreatic tissue in the region conforming to the solid area, submucosa lined with intestinal mucosa separated by a smooth border, and a cyst wall containing muscle tissue were observed in Hematoxylin-eosin (H&E) staining (c). The histochemical examination showed that the surface epithelium contains intestinal type mucin with PAS-AB (d)

Pathology report:

In the microscopic evaluation, pancreatic tissue in the region conforming to the solid area, submucosa lined with intestinal mucosa separated by a smooth border, and a cyst wall containing muscle tissue were observed (Figure 4c). The histochemical examination showed that the surface epithelium contains intestinal type mucin with PAS-AB (Figure 4d). In the light of histopathological evaluation, the case was reported as intestinal duplication cyst located in the pancreas.

Her general condition improved in the postoperative follow-up. Afterwards, she did not have pancreatitis attacks again during her hospitalization and in the 8-month follow-up in postoperative period. Atrophic changes in the pancreas and enlargement of the Wirsung duct were found in the MRI examination performed at the postoperative 6th month.

Discussion:

Pancreatitis is an uncommon cause of acute abdomen in children. In recent years, with the increase in obesity, pancreatitis is also seen at increasing rates. The increase in the incidence of obesity, which is one of the most common causes of pancreatitis in children, is shown as the reason for this [3].

Fifteen percent of cases of acute pancreatitis in children are caused by structural anomalies as congenital anomalies (e.g., pancreas divisum), Crohn's disease, and duodenal ulcers that involve the periampullary region [4].

There are very few duodenal duplication cyst described in the literature account for 5 – 10 % of gastrointestinal duplications with an incidence of less than 1 per 100 000 births. Chen et al. reviewed 47 cases of duodenal duplication cysts [5]. Hunter et al. Reviewed 43 cases of enteric duplication cysts were reported in association with the pancreas. Duplication cysts are tubular or cystic formations containing gastrointestinal mucosa surrounded by a muscle layer.

An important and very rare feature is that between 25 and 35% of duplication cysts may contain ectopic gastric mucosa leading complications as bleeding . Mostly of them associated with some portion of the gastrointestinal tract [6]. In rare cases cysts can be isolated and have a separate vascular pedicle Isolated retroperitoneal enteric duplication cysts are also in this group [7].

In our case, a 12-year-old female patient had a peristaltic and hemorrhagic cystic lesion located in the head of pancreas, with no intestinal connection, and containing duodenal mucosa surrounded by a muscle layer.

It is possible to distinguish intra-pancreatic duplication cysts from pseudocysts by selecting the wall structure in a good quality US examination using the fluid-filled gastric acoustic window. At the same time, peristatilism can also be evaluated sonographically, since it is a real-time examination.

CT and MRI examinations are useful in identifying the location and size of the cyst as well as accompanying lesions. In addition, MRCP can be used to identify the relationship of the lesion with the pancreatic duct and other ductal anomalies that may accompany it.

Conclusion:

Congenital anomalies and variations should be kept in mind when evaluating diseases in the pediatric patient group. In many diseases, especially in children, real-time sonographic evaluations shed light on the path to the correct diagnosis.

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