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A 7-Year-Old Girl with Early Developed Pseudocyst After Acute Pancreatitis: A Case Report and Literature Review



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KEYWORDS pancreatic pseudocyst, pancreatitis, pediatrics, ultrasonography Abstract We report a 7-year-old Taiwanese girl with acute pancreatitis (AP) complicated with pseudocyst (PC). The girl was found to have a PC by ultrasonograpgy (USG) and magnetic resonance imaging 14 days after the onset of AP. The girl was discharged 21 days after manage- ment with an asymptomatic PC. The diagnostic criteria of AP included abdominal pain, serum amylase or lipase level, and imaging findings. Transabdominal USG after appropriate prepara- tion with adequate fasting, intake of some liquids during the scanning, and right decubitus po- sition enhance the demonstration of pancreas and peripancreatic structures. PC could be seen in up to 38% of pediatric AP patients. It can form within 2 weeks after the onset of symptoms, although most are late complications. Pancreatic PCs have to be differentiated from other intra-abdominal cysts on USG according to their image character and anatomic location. A well-prepared USG examination in combination with liquid intake and right decubitus position is of value in the diagnosis and follow-up of PC. © 2017, Elsevier Taiwan LLC and the Chinese Taipei Society of Ultrasound in Medicine. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/ by-nc-nd/4.0/).		
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Abbreviations: ANC, acute necrotic collection; AP, acute pancreatitis; APFC, acute peripancreatic fluid collection; CECT, contrastenhanced computed tomography; CRP, C-reactive protein; CT, computed tomography; MRI, magnetic resonance imaging; PAP, pediatric acute pancreatitis; PC, pseudocyst; USG, ultrasonography.

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Introduction

Acute pancreatitis (AP) in children has gained more attention in the past decades, and an increase in incidence was documented [1-3]. Pseudocysts (PCs), which are a wellknown late complication of AP, mostly take 4 weeks to develop, but can also encapsulate earlier [4]. Transabdominal ultrasonography (USG), rather than computed tomography (CT), is commonly used as the first-line image study tool in pediatric acute pancreatitis (PAP) patients because of its radiation-free property. It is challenging to overcome the limitations of USG in detecting the pancreas and peripancreatic structures.

We present the case of a 7-year-old Taiwanese girl with AP who developed PC 14 days after the onset of symptoms. The literature on PAP and PC was reviewed, with special focus on USG imaging studies.

Case Report

This 7-year-old previously healthy Taiwanese girl presented to our emergency department with upper periumbilical pain and bilious vomiting for 2 days. The pain was first noticed when she woke up from her sleep at midnight. The pain radiated to the back and was relieved by bending the body. Fever was recorded once 2 days ago (with temperature up to 38.9° C). Abdominal trauma history, travel or contact history, alcohol or medicine use, and family history of hyperlipidemia or pancreatitis were all denied. She had been to another hospital earlier on the same day, where the diagnosis of AP was made based on the symptoms, with elevated amylase and lipase (220.5 U/L and 150 U/L, respectively) and interstitial edematous pancreas with peripancreatic fluid collections demonstrated by the abdominal CT (Figure 1).

The initial physical examination showed that the girl was anicteric and restless on the bed. Her body weight was 42.5 kg with a body mass index of 22.6 kg/m². The following vital signs were noted: body temperature, 37.7° C; heart rate, 123 beats/min; respiratory rate, 24 times/min; blood pressure, 108/22 mmHg. Upper periumbilical and left upper quadrant tenderness were noticed without muscle guarding, rebounding, or palpable mass. Transabdominal USG reported swelling and blurred margin of the pancreatic body (diameter 1.6 cm) and pyoascites at the splenorenal recess (Figure 1). The biliary system appeared normal. Her laboratory data supported the diagnosis of AP (Table 1).

Adequate intravenous hydration, pain control with morphine, and gabexate mesylate were administered. Antibiotics were ordered because intra-abdominal infection could not be ruled out. Parenteral nutrition and nasogastric tube for drainage were provided. Oral feeding was started since the 11th hospital day.

Follow-up abdominal USG 6 days after the previous examination showed less swelling of pancreatic body. However, the pancreatic tail could not be demonstrated because of the presence of too much gas. Abdominal magnetic resonance imaging (MRI) done on the 12^{th} hospital day (14 days after the onset of abdominal pain) showed a 6 cm \times 4-cm infectious PC near the pancreatic tail and inflammatory ascites at the splenorenal recess (Figure 2).

The PC was followed up by USG on the 15th and 20th hospital days, and the size was 6.9 cm \times 4.6 cm and 7.2 cm \times 3.6 cm, respectively (Figure 3).

The lipase level mildly rebounded to 78 U/L after 5 days of enteral feeding, but there was no recurrence of abdominal pain or vomiting. The serum triglyceride level dropped to 454 mg/dL, and total cholesterol dropped to 163 mg/dL in 8 days without medication use. After adequate oral intake could be maintained, the girl was discharged 21 days after management.

Discussion

PAP was defined clinically by the INSPPIRE (International Study group of Pediatric Pancreatitis: In search for a cure) as the presence of at least two of the three following criteria: (1) abdominal pain compatible with acute AP; (2) serum amylase and/or lipase values at least 3 times greater than the upper limits of normal; and (3) imaging findings characteristic of AP [5]. The disease incidence is estimated to be 2.4-13.2 per 100,000 children, with an increasing trend across all age groups [2].

The most common etiology of PAP include trauma and systemic disease. Yeung et al [6] reported that one-third (16 out of 43) of AP cases were attributable to trauma, of whom 50% required surgery [3]. A more recent study reported that 37% of PAP cases were idiopathic, followed by drugs, gallstones, hereditary, and organic acidemias [7]. The complications of AP were divided as local or systemic according to the revision of the Atlanta classification published in 2012 [8]. The early phase of local complication is further classified into two distinct morphologic features delineated by abdominal contrast-enhanced CT (CECT) or contrast-enhanced MRI: acute peripancreatic fluid collection (APFC) and acute necrotic collection (ANC). A radiologically identifiable capsule formed 4 weeks after the onset of symptoms is the landmark feature of late-phase local complication. PC progresses in half of the patients with APFC, and walled-off necrosis is formed after the ANC becomes organized [9].

PCs are best detected by CECT. A well-circumscribed, usually round or oval peripancreatic fluid collections of homogeneously low attenuation that are surrounded by a well-defined enhancing wall consisting of fibrous or granulation tissue is diagnostic of PC. The enhancing capsule rarely develops earlier than 4 weeks after the onset of AP [4]. On MRI, PCs are detected by T2-weighted sequences without fat saturation as bright fluid signal intensity. When contrast-enhanced MRI is used, the rim of the PC can be evaluated for thickening and enhancement, which suggests superimposed infection [10]. Anatomically, most extrapancreatic PCs were located in the body and tail region, whereas most intrapancreatic PCs were in the head of the pancreas [11]. In one recent study that included 787 adult patients with pancreatic PCs larger than 5 cm, 64.3% were located at the head, neck, or body of pancreas, 29.9% at the tail, and the rest at the uncinate [12].

Transabdominal USG has 75% to 90% sensitivity rates in the detection of pancreatic PCs, which is inferior to those of CT, owing to the presence of the overlying bowel gas. PCs appear on USG as a well-defined, round or oval anechoic



Figure 1 Contrast enhanced computed tomographic (CECT) and ultrasonography (USG) of abdomen done on the day of admission. (A) Horizontal and (B) coronal section showed that the pancreas was edematous with peripancreatic fluid collections (arrowheads). (C) Swelling and blurred margin of the pancreatic body, 1.6 cm in diameter. (D) Hypoechoic area with heterogeneous content between the spleen and the left kidney, indicating pyoascites at splenorenal recess (arrow). LK = left kidney; P = pancreas; S = spleen.

structure contained within a smooth wall associated with distal acoustic enhancement. It can have a more complex appearance, with varying degrees of internal echoes resulting from the presence of necrotic debris in an earlier

Table 1Laboratory data.			
Variable	On admission	Reference range ^a	
Hemoglobin (g/dL)	12.5	11.5–14.5	
Leukocytes (/µL)	13,400	4000-12,000	
Segment (%)	78.6	55—75	
Platelet (/µL)	198,000	140,000-450,000	
Amylase (U/L)	168	26–115	
Lipase (U/L)	125	22–51	
C-reactive protein (mg/dL)	24.4	0.05-1.0	
Glucose (mg/dL)	96	60-100	
Triglyceride (mg/dL)	601	35—150	
Cholesterol (mg/dL)	247	130-200	

^a In cases where pediatric reference range is provided, the range for a 7-year-old female is applied. Otherwise, the reference range for an adult used in MacKay Memorial Hospital is used. phase, when hemorrhage occurs into the cyst, or when infection of the cyst complicates the clinical course [13]. USG is commonly used as the first-line image study tool in PAP patients because of its radiation-free, hands-on, and repeatable properties. Its usefulness is attributable to the optimal sonic window provided by the relative lack of fat tissue and the large left hepatic lobe in children. A 5.0- or 7.5-MHz sector transducer is usually used in USG evaluation of children with suspected pancreatic disease [14]. Although noncooperation of young children with USG examinations is a frequently encountered problem, appropriate preparation prior to the examination with adequate fasting for 6-8 hours in children and 3 hours in neonates, and intake of some liquids during the scanning, with or without right decubitus position, both promote the demonstration of pancreas and peripancreatic structures.

Pancreatic PCs have to be differentiated from other intra-abdominal cysts on USG according to its image character and anatomic location. Histopathologically, intraabdominal cysts include six groups: (1) cysts of lymphatic origin; (2) cysts of mesothelial origin; (3) enteric cysts; (4) cysts of urogenital origin; (5) dermoid cysts; and (6) PCs—infectious or traumatic etiology [15]. Pancreatic cysts can be secondary to neoplasms with high malignancy rate such as intraductal papillary mucinous neoplasms, or solely



Figure 2 Magnetic resonance imaging (MRI) of abdomen done 14 days after the onset of abdominal pain. A 6 cm \times 4-cm welldefined cystic lesion near the pancreas tail with high signal in (A) T2-weighted image (T2WI) and (B) low signal in apparent diffusion coefficient sequence (ADC) (arrowhead), indicating infectious pseudocyst. (C) Focal high signal in T2WI and low signal in ADC (D) at left splenorenal recess, favoring inflammatory ascites (arrows). C = pseudocyst; G = stomach; LK = left kidney; S = spleen.



Figure 3 Ultrasonography (USG) of abdomen. (A) On the 15^{th} hospital day, a 6.9 cm \times 4.6-cm pseudocyst at the pancreatic tail with immature capsule except the wall close to the spleen. (B) On the 20^{th} hospital day, the 7.2 cm \times 3.6-cm pseudocyst was identified after drinking a cup of water and right decubitus position. Note the capsule had become more concrete. C = pseudocyst; G = stomach; S = spleen.

benign on the other end of the spectrum. Pancreatic cysts with sizes smaller than 3 cm, no solid component, and no main pancreatic duct dilatation are at low risk for malignancy and could be managed with surveillance only [16]. In contrast to "true" cysts, PCs lack epithelial lining over the inner wall of the fibrous capsule [17]. Choledochal cyst is characterized by dilation of the common bile duct with associated intrahepatic ductal dilation [18]. One should trace the continuity between the cystic mass and the biliary tract to make the diagnosis of choledochal cyst. Retroperitoneal, omental, and mesenteric cysts are very rare intra-abdominal tumors derived from ectopic lymphatic tissue, and preoperative localization of cysts by either USG or CT was possible only in three out of 23 patients in one study [19]. Typical USG findings of a cyst include round shape, smooth wall, absence of internal echoes, presence of lateral shadowing, and posterior echo enhancement. Retroperitoneal cysts, by contrast, are usually imaged as a round mass behind the pancreas, with dense internal echoes without lateral shadowing or posterior echo enhancement, which presents a "pseudo-solid" pattern [20].

Pancreatic PC in children was believed to be a rare condition because most series described only a small number of patients [21–23]. However, some recent studies may challenge this concept. Bolia et al [24] reported that 38% of PAP patients developed PCs, and half of them required drainage. Poddar et al [1] retrospectively reviewed 320 pediatric pancreatitis patients, including 160 patients with AP, 67 with acute recurrent pancreatitis, and 93 with chronic pancreatitis, and found that 60 patients (18.7%) developed PCs. History of abdominal trauma was documented in 27.2% to 100% of children with PC, but whether traumatic or nontraumatic PCs are more likely to resolve spontaneously is conflicting [21-24]. Putting together nine studies mentioning pediatric PCs, a total of 212 cases were reported [1,6,21-27]. Among the 152 cases with predisposing factor reported, 112 (73.7%) had a history of abdominal trauma and 24 (15.8%) were idiopathic. Of the 133 cases assessed for symptoms, 61 (45.9%) had abdominal pain, 45 (33.8%) had vomiting, and 19 (14.3%) had palpable mass. Thirty-three patients had symptoms not further specified. The management showed substantial diversity: 58 (27.4%) patients underwent internal drainage either with cystgastrostomy or cystjejunostomy, 49 (23.1%) patients underwent external drainage, and 58 (27.4%) patients were treated conservatively.

It is recommended that PCs be drained if either symptoms or complications developed, regardless of the size [1]. More than half of the PCs presenting in children required drainage, ranging from 50% to 70.8% [1,21,23,24]. Neither symptom nor complication was found during the follow-up period including repeated USG in this case; thus, drainage was withheld.

In conclusion, PC is not an uncommon complication following PAP, and can form within 2 weeks after the onset of symptoms. CECT is the best modality to detect PC, and a well-prepared USG examination in combination with liquid intake and right decubitus position is of value in the diagnosis and follow-up of PC. PCs could be managed expectantly regardless of the size, unless symptoms or complications develop. References

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