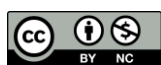


Case Report

Diagnostic and Management Approach of Pancreatic Pseudocyst in Children

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Abstract:

Background: Pancreatic pseudocyst is a fluid-filled sac within the pancreas encapsulated by fibrous tissue. Blunt-abdominal trauma is the leading risk factor in children. Due to varied and non-specific clinical manifestations, diagnosis can be challenging. Thus, this study aimed to explain the diagnostic and management approach of pediatric pancreatic pseudocyst.

Case: An 8-year-old boy presented with a 3-month history of progressive abdominal mass accompanied by abdominal pain, bilious vomiting, constipation, and weight loss. Physical examination revealed a skin-colored mass located in the epigastric region. A CT-scan confirmed a cystic lesion with well-defined borders in the pancreas. Laboratory tests indicated elevated levels of plasma amylase and lipase enzymes. The patient underwent endoscopic ultrasound (EUS), followed by cyst drainage. Analysis of the pseudocyst fluid revealed increased amylase and lipase enzymes, and carbohydrate antigen 19-9 (CA 19-9) levels.

Discussion: A thorough patient history and physical examination are essential in diagnosing pancreatic pseudocyst. While CT-scan provides valuable information, EUS has higher sensitivity and specificity for diagnosis. Amylase and lipase enzymes levels are frequently elevated, and CA-19-9 can be useful, however, should be complemented with other biomarkers. Drainage is indicated for cysts that do not resolve spontaneously. Adequate nutrition is also crucial for successful patient management.

Conclusion: Pancreatic pseudocysts should be considered in children with an abdominal mass following blunt-abdominal trauma. Endoscopic ultrasound (EUS) is a valuable tool for both diagnosing and assisting the management of pancreatic pseudocysts.

Keywords: blunt abdominal trauma, endoscopic ultrasound, pancreatic pseudocysts

Introduction

Pancreatic pseudocyst is defined as a fluid-filled sac within the pancreas that contains pancreatic enzymes and necrotic tissues. It is surrounded by a non-epithelial fibrous tissue.^{1,2} The incidence of pancreatic pseudocyst is low, accounting for 1.6 to 4.5% per year. Currently, comprehensive data on pancreatic pseudocyst in pediatric is still limited due to its rare occurrence in children.³

Blunt abdominal trauma is the most common risk factor of pediatric pancreatic pseudocyst.³⁻⁶ Pancreatic pseudocyst is more common in boys with an average age of onset around 7.5 years old.¹ Clinical presentations are often varied and typically exhibited as non-specific gastrointestinal symptoms such as abdominal pain, vomiting, abdominal mass, and fever. Endoscopic ultrasound (EUS) accompanied with drainage is the current first-line treatment for pancreatic pseudocyst.^{3,7}

Despite being rare in children, pancreatic pseudocyst has high morbidity if left untreated.⁵ Previous studies have reported that 30 – 50% of persistent, untreated pseudocyst could lead to complications, such as abscess formation, fistula, spontaneous rupture, and massive bleeding, which may result in death.⁸ Pancreatic pseudocyst larger than 6 centimeters that shows no improvement after 6 weeks generally requires medical intervention. This establishes the importance of early identification and treatment to prevent unwanted complications. Thus, this study aims to explain the diagnostic and management approach for pancreatic pseudocyst in children due to blunt abdominal trauma.

Case

An 8-year-old boy was referred to the tertiary, national-referral hospital with a chief complaint of abdominal mass that had been present for three-months prior to admission. Four months prior to admission, he and his mother had a motorcycle accident. During the accident, he fell from the motorcycle and got hit in the stomach by the motorcycle handle. Subsequently, the patient experienced intermittent abdominal pain in the upper left quadrant and the epigastric region, with a 3-4 on a Visual Acuity Score (VAS). The pain was subsided with analgesic.

Three months prior to the admission, the patient had another abdominal trauma as he was punched on the left side of his stomach during a playfight with his friend. After the incident, he started to experience nausea and vomiting 2-4 times per day, consisting of food. Then, he went to a primary healthcare center and was given anti-nausea medication. However, the symptom only mildly decreased and continued to appear intermittently. He also started to notice a growing, skin-colored lump in the abdomen at the size of a chicken egg, with no redness and no bruising. He experienced intermittent abdominal pain, with an intensity of VAS 4-5, localized in the same area

as the initial injury. The pain was unaffected by position or food intake and did not radiate to the back or chest. There was no fever, breathing difficulty, icterus, jaundice, or changes in urination or defecation. Despite maintaining a good appetite, he experienced an unintended weight loss of 4 kg over the past three months.

Three weeks prior to hospital admission, he experienced a recurrence of nausea and vomiting. The vomit was bilious, occurring 2-4 times per day. Furthermore, the lump has reportedly grown progressively over the past month. He also suffered more frequent, severe abdominal pain. In addition, he was constipated for three days. Due to these worsening symptoms, he was admitted to the regional public hospital and underwent an abdominal CT-scan with contrast for further evaluation. The scan revealed a 13.5 x 11.36 x 17.03 cm cystic lesion with well-defined borders and a lobulated margin in the corpus pancreas, suggestive of a pancreas pseudocyst (Figure 1). No abnormalities were found in other organs. The patient was prescribed anti-nausea and laxative for 5 days, after which the symptoms subsided. The patient was then referred to Dr. Cipto Mangunkusumo General Hospital (RSCM) for further evaluation of the suspected pancreas pseudocyst.

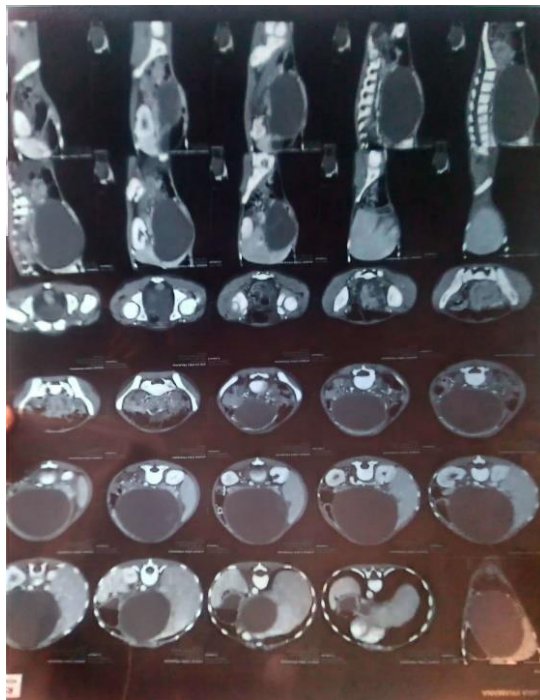


Figure 1. Abdominal CT-scan with contrast

The day before admission to RSCM, he experienced three episodes of non-bilious vomit in 24 hours and new episode of abdominal pain with a VAS of 2-3. There was no fever, constipation, dyspnea, cough, or coryza. Upon admission to RSCM, he was no longer experiencing nausea, and his oral intake was sufficient.

On physical examination, he looked weighed 17.6 kg, was 123 cm tall, and had an upper arm circumference of 12 cm. This indicates malnutrition (based on upper arm circumference) with severely underweight, but normal stature. His vital sign was within normal limits. Abdominal physical examination revealed a distended abdomen with normal bowel sounds (Figure 2). A 21 cm x 15 cm well-defined, skin-colored mass was palpated in the epigastric region. The liver was difficult to assess, and the spleen was not palpable. No other abnormalities were found during the physical examination.

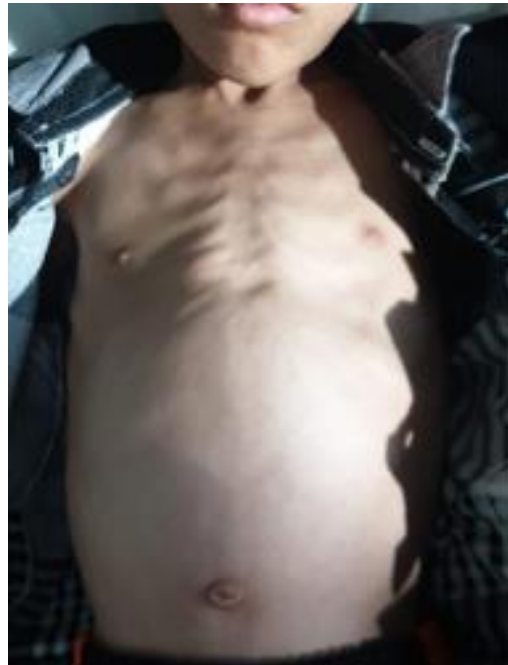


Figure 2. Patient clinical appearance

Laboratory tests indicated normocytic normochromic anaemia (Hb 10.8 g/dL, MCV 79.9 fL, MCH 26.2 pg, MCHC 32.8 g/dL), with normal leucocyte and thrombocyte counts. Additionally, the patient exhibited a mild hypokalaemia, hypophosphatemia, and increased of amylase and lipase enzymes levels (3.2 mEq/L, 3.6 mg/dL, 160 U/L and 343 U/L, respectively). Plasma albumin (3,8 g/dL), AST/ALT (14 U/L / 26 U/L), ureum (30 mg/dL), creatinine (0,4 mg/dL), and PT/APTT (1x/1.3x) levels remained within normal range.

On the sixth day of hospitalization, the patient was scheduled to underwent endoscopic ultrasound (EUS) and esophagogastroduodenoscopy (EGD). The EUS revealed a well-defined, 11 cm x 10 cm anechoic cyst pressing against the anterior wall of the stomach. The EGD examination showed a grade B esophagitis accompanied by gastritis, delayed gastric emptying, and a Forrest class III ulcer in the corpus, as well as laryngopharyngeal reflux. The EUS examination was then followed by the drainage of the pancreatic pseudocyst using a double pigtail plastic stent.

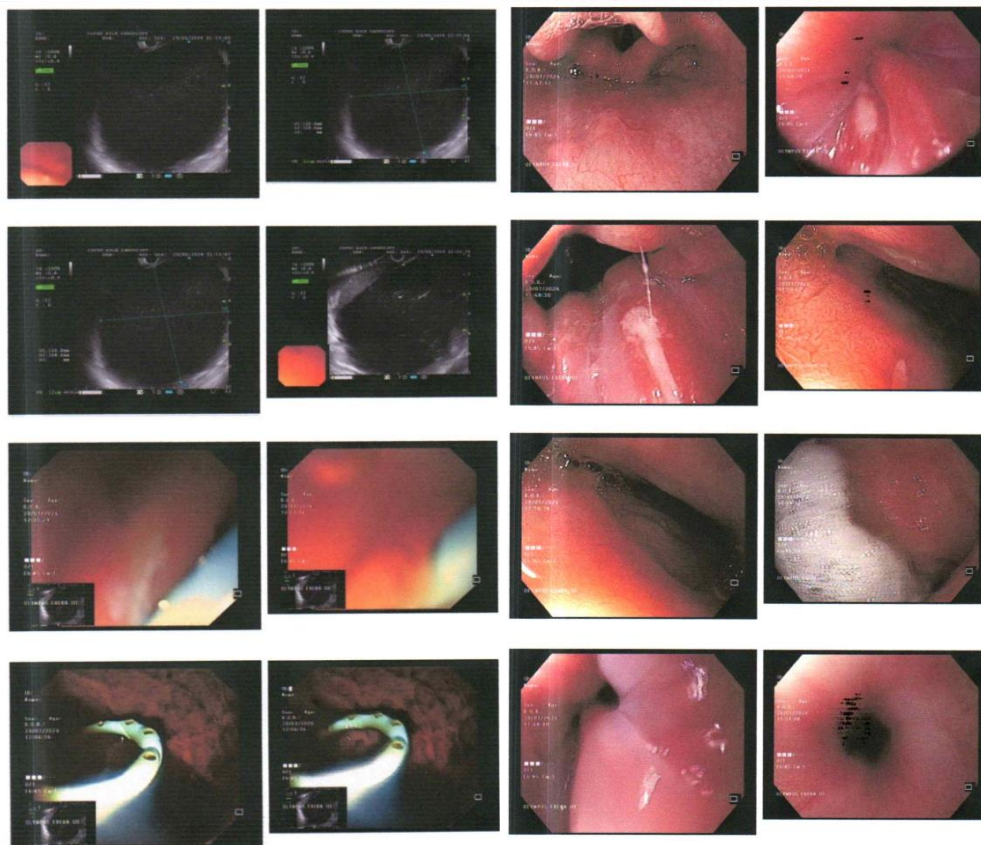


Figure 3. Endoscopic Ultrasound (EUS) and Esofagastroduodenoskopi (EGD)

After drainage, the patient received total parental nutrition with a total fluid intake of 1300 ml/24 hours and an initial glucose infusion rate of 6 mg/kg/min. Gradually, he was transitioned to oral intake, starting with 5 x 250 ml of specialized formula and one regular meal per day (lunch, 300 kcal). Patient received intravenous proton pump inhibitor (PPI) esomeprazole 20 mg every 12 hours for 3 days, then continued with intravenous omeprazole 20 mg twice a day. The administration of omeprazole 20 mg every 12 hours is planned to be continued orally before meals for 4 weeks, and then, will be reduced to 20 mg every 24 hours orally for 8 weeks.

The result of pseudocyst fluid analysis showed elevated levels of amylase, lipase, as well as carbohydrate antigen 19-9 (CA 19-9) (8583 U/L, 271270 U/L, and 2711.9 U/mL, respectively). Conversely, a carcinoembryonic antigen (CEA) result remained low (CEA < 0.5 ng/mL). On the seventh day of treatment, the plasma amylase enzyme level (60 U/L) remained elevated above the normal range (<31 U/L) but showed improvement compared to previous results. Meanwhile, the lipase enzyme level (62 U/L) had decreased to within the normal range. The patient was subsequently discharged after becoming symptom-free. Monthly follow-ups showed he was no longer exhibited any complaints and had improved nutritional status.

Discussion

This case report describes an 8-year boy who presented with a three-month history of abdominal mass. This mass was developed after two episodes of blunt abdominal trauma, four months prior and the other three month prior to hospitalization. The patient also experienced recurrent abdominal pain, nausea, bilious vomiting, weight loss, and constipation.

Several differential diagnoses could be considered in this case. Due to the absence of splenomegaly on physical examination, the possibility of a mass in the spleen can be ruled out. Additionally, the absence of urinary disturbances or high blood pressure made kidney malignancy or hydronephrosis unlikely. These differential diagnoses were further confirmed by normal findings of spleen and kidney in abdominal CT scan.

Furthermore, several potential diagnoses for this patient's abdominal pain could be considered, including functional gastrointestinal disorder (FGID), such as gastroesophageal reflux disease (GERD), irritable bowel syndrome (IBS), or functional dyspepsia. However, based on the characteristics of the pain, which were not affected by food or drink intake, and the presence of an abdominal mass preceded by a clear history of trauma in the patient made FGID an unlikely primary cause for the patient's complaints. The patient's symptoms of constipation and bilious vomiting suggest an obstruction in the gastrointestinal tract. This is similar to a case report of an 8-year-old patient with a pancreatic pseudocyst reported by Suleman et al.⁹ Common symptoms that are found in patients with pancreatic pseudocyst include abdominal pain (76-90%), nausea and vomiting (50%), and weight loss (20-51%)¹⁰, all of which is shown in this patient.

The blunt abdominal trauma in this patient is likely caused an injury to the pancreatic duct, leading to the extravasation of pancreatic fluid. Subsequently, this fluid formed a localized sac surrounded by the walls of adjacent organs, such as the pancreas, omentum, and colon. Furthermore, pseudocysts generally take 4-6 weeks to develop.⁴ This timeframe aligns with the onset of the patient's abdominal mass symptoms. Imaging studies are the most important diagnostic modality for establishing the diagnosis of pancreatic cystic lesions, such as pancreatic pseudocyst. Abdominal ultrasound is often used to evaluate suspected pseudocysts, typically showing an anechoic round or oval structure with distal acoustic enhancement. CT scans can also evaluate pseudocysts and identify other surrounding pathologies, but may have difficulty to differentiate pseudocysts from neoplasms. On CT scans, pancreatic pseudocyst appears as well-defined, low-attenuation, and homogeneous. Magnetic Resonance Imaging (MRI) is the most sensitive and accurate method for diagnosing pancreatic pseudocyst, but it is not routinely used in clinical practice as CT scans usually already provide sufficient diagnostic information.

Endoscopic ultrasound (EUS) is another diagnostic tool for pancreatic pseudocyst, which has high sensitivity and specificity (93-100% and 92-98%, respectively). This makes it more superior than CT scans, which only have a sensitivity of 90-100%.¹¹ EUS also avoids radiation exposure and can differentiate pseudocyst and other pancreatic cyst.^{3,7} While endoscopic retrograde cholangio-pancreatography (ERCP) and magnetic resonance cholangiopancreatography (MRCP) can also contribute to the diagnosis of pancreatic pseudocyst, their availability is significantly limited in developing countries.³ In this patient, a CT scan initially identified a pancreatic pseudocyst and for further evaluation, the patient underwent EUS.

Several laboratory markers are useful in diagnosing pancreatic pseudocyst. CA 19-9, a polymer glycoprotein used as a biomarker for pancreatic malignancy, is not specific for detecting malignancy since it can also be elevated in various conditions, such as pancreatitis, pancreatic cysts, diabetes mellitus, liver fibrosis, and cholestasis.^{12, 13} CA 19-9 levels below 37 U/mL suggest pseudocyst or serous cystadenoma over other types of pancreatic cystic lesions. CA 19-9 shows high specificity (98%) but low sensitivity (19%).^{12, 14} Therefore, CA 19-9 should be combined with other biomarkers, particularly amylase and lipase. A meta-analysis found that an amylase concentration of <250 U/L in cyst fluid has excellent specificity (98%) for ruling out the diagnosis of pseudocyst.¹⁵ Another study revealed that amylase levels >8500 U/L were observed in 91% of pseudocyst cases.¹⁶ Another study showed that amylase levels >5000 U/L were found in 94% of cases, and lipase levels >2000 U/L were found in all cases of pseudocyst.¹⁷

In other diagnoses, such as mucinous cystic neoplasm and mucinous cystadenocarcinoma, amylase and lipase levels are typically low, while other tumor markers, such as CEA and CA 125, may be elevated.³ In this case, the patient had elevated blood amylase and lipase levels, as well as elevated amylase, lipase, and CA 19-9, levels from the cyst fluid analysis. However, CEA levels remained low. These findings support the diagnosis of a pancreatic pseudocyst.

Drainage was indicated in this case as the pseudocyst had persisted for over 6 weeks and exceeded 6 cm in size, making spontaneous resolution unlikely.¹⁸ Ultrasound and endoscopy were used to guide the drainage procedure, allowing identification of vascular structures around the cyst. Studies show EUS-guided drainage has a success rate of 78-89% and a complication rate of 4-7%, making it a safer option than conventional drainage.⁴ In addition to EUS-guided drainage, surgical drainage may be indicated for pancreatic pseudocysts with complications, such as infection, necrosis, duct strictures, biliary stenosis, or compression of adjacent structures.²

Fully-covered self-expanding metal stents (FCSEMS) are an alternative to plastic stent, offering a lower risk of occlusion and potentially reducing the need for repeat procedures. While there is still ongoing debate regarding the pros and cons of FCSEMS versus plastic stents in pseudocyst drainage, a study by Sharaiha et al. reported that plastic stents have a 2.5 times higher risk of complication.¹⁹ However, a meta-analysis by Saunders, which included 698 patients, revealed no significant differences in success rates, complications, and recurrence rates between FCSEMS and plastic stents.²⁰ In this case, a double pigtail plastic stent was used, and the clinical outcome was favourable.

Adequate nutrition is crucial in managing pancreatic cystic lesions. The optimal timing for initiating enteral nutrition in patients with pancreatic pseudocysts remains a subject of ongoing debate. However, an early initiation of enteral feeding within 48 hours in patients with severe acute pancreatitis is associated with lower incidence of infection, shorter hospital stays, and decreased mortality rates compared to delayed enteral feeding or total parenteral nutrition.²¹ In this case, the patient received enteral nutrition prior to the procedural intervention, followed by total parenteral nutrition post-procedure, and gradually transitioned to enteral nutrition. The patient showed a weight gain of 3.85 kg, increasing from 16.9 kg to 20.75 kg in one month. Notably, the enteral nutrition provided to the patient included a formula comprising 50% medium-chain triglycerides (MCT). To date, there are no randomized controlled trials assessing the efficacy of different nutritional formulas, including both standard and MCT-enriched formulas.²² Nevertheless, using a predigested enteral formula containing a combination of long-chain fatty acids and MCTs, as used in this case, may be beneficial. MCTs require less reliance on lipase activity for their absorption, thereby facilitating easier digestion.²³

The prognosis for the patient is generally good, as clinical improvement has already been observed following treatment. The definitive therapy by endoscopic-assisted drainage has an almost 100% success rate in managing pseudocysts.²⁴

Conclusion

Pancreatic pseudocyst should be considered in children presenting with abdominal mass and a history of blunt abdominal trauma. While CT imaging provides valuable diagnostic information, EUS offers the highest sensitivity and specificity for confirming the diagnosis. Fluid analysis from the cyst gives more accurate diagnostic data compared to serum analysis. The management of guidance drainage as well as adequate nutritional support, has been associated with favorable outcomes.

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Conflict of Interest

None declared

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