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Case Report

Pancreatic Stones in Children with Chronic Pancreatitis: A Case Report

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Abstract:

Background: Chronic pancreatitis is a continuous progressive inflammation of the pancreas that can cause recurrent abdominal pain. The incidence of chronic pancreatitis in children is on an upward trend in recent years. The purpose of this case report is to enhance clinician's insight in considering chronic pancreatitis as a differential diagnosis in cases of recurrent abdominal pain in children.

Case: We reported a case of chronic pancreatitis with pancreatic stones in a 12-year-old child. The patient came with complaints of recurrent upper left abdominal and heartburn accompanied by nausea and vomiting. Epigastric and left hypochondriac tenderness were found. Laboratory examination showed an increase in pancreatic enzymes up to 6x the normal value. The results of abdominal CT-scan and MRCP showed multiple pancreatic stones with pseudocysts. The patient then underwent the Puestow procedure. Postoperatively, the complaints of abdominal pain resolved and the pancreatic enzymes improved significantly.

Discussion: The diagnosis of pancreatitis must meet 2 out of the 3 criteria that have been determined. Primary management includes fluids, analgesics, early nutrition, and surgery in cases of chronic pancreatitis accompanied by complications such as pseudocysts or pancreatic stones.

Conclusion: This case teaches the need for a careful clinical approach and consideration of chronic pancreatitis as a differential diagnosis in a child with recurrent abdominal pain.

Keywords: abdominal pain, chronic pancreatitis, pancreatic stone, children

Introduction

Pancreatitis is an inflammatory process of the pancreas. This disease is often overlooked as a cause of abdominal pain in children so it is rarely diagnosed (underdiagnosed disease). Pancreatitis is divided into 3 categories, namely acute pancreatitis, acute recurrent pancreatitis and chronic pancreatitis. In the last 10 years, the incidence of acute pancreatitis in children has increased by 3.6-13.3 cases per 100,000 children.[1] One study estimated that 9-35% of children with acute pancreatitis

will have recurrent attacks.[2] The incidence of chronic pancreatitis also continues to rise, with an estimation of 2 cases per 100,000 children per year.[3] Chronic pancreatitis is a progressive inflammation that causes damage to the morphology and structure of the pancreatic parenchyma, hence negatively affecting pancreatic functions. The diagnosis of chronic pancreatitis is established when abdominal pain is consistent with the location of the pancreas and radiological findings suggest chronic pancreatic damage, evidence of exocrine and endocrine insufficiency of the pancreas, or biopsy results show histopathological abnormalities consistent with chronic pancreatitis.[4] Pancreatic calculi is a late complication of chronic pancreatitis regardless of the etiology, that can appear in the ducts, branches or pancreatic parenchyma.[5]

Case

A 12-year-old boy was admitted to the emergency room with chief complaint of epigastric abdominal pain and left upper abdomen radiating to the back in the past day. Pain was episodic with sensation of being stabbed and VAS 8. Complaints were accompanied by nausea and vomiting. History of jaundice was denied. Patient did not have any fever and urination as well as bowel movements were normal. Similar complaints were felt in the last 9 months, which the pain resolved with the administration of analgesics and then relapsed 2-3 months later. Physical examination revealed epigastric and left hypochondriac tenderness. Laboratory examinations were carried out with the results of an increase in pancreatic amylase by 6x the normal value (amylase 327 u/L) and pancreatic lipase by 3x the normal value (lipase 176 u/L). No hemoconcentration, leukocytosis, or electrolyte abnormalities were found. Liver, kidney, bilirubin and albumin functions were within normal limits.

Abdominal CT scan with contrast showed multiple stones and prominent calcification of the pancreatic parenchyma with dilated pancreatic ducts and multiple intraductal stones in it, indicating a chronic pancreatitis process. (**Figure 1** and **Figure 2**)

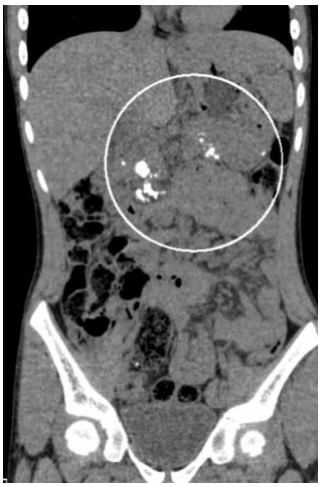

Figure 1. Coronal view of abdominal CT scan with contrast showing multiple pancreatic stones (circle mark).

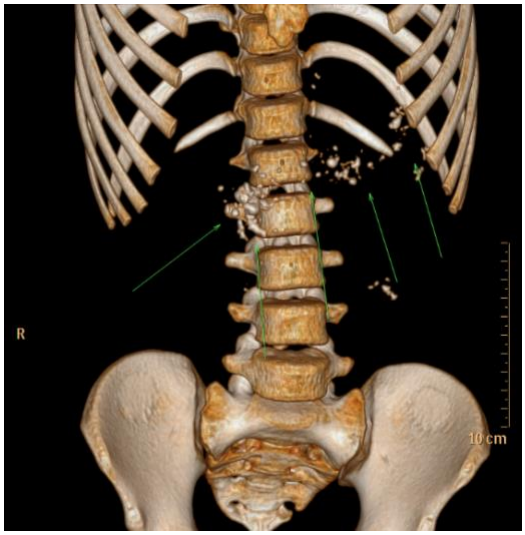

Figure 2. Three-dimensional visualization of abdominal CT scan showing multiple stones (green arrows) in the pancreas.

Magnetic Resonance Cholangiopancreatography (MRCP) showed dilatation of the pancreatic ducts due to multiple stones in the head and body of the pancreas with the largest diameter being 2.3 cm as well as multiple peripancreatic pseudocysts with the largest diameter being 4 cm with internal stone components. This description is consistent with chronic pancreatitis with pancreatic stones. (**Figure 3**)

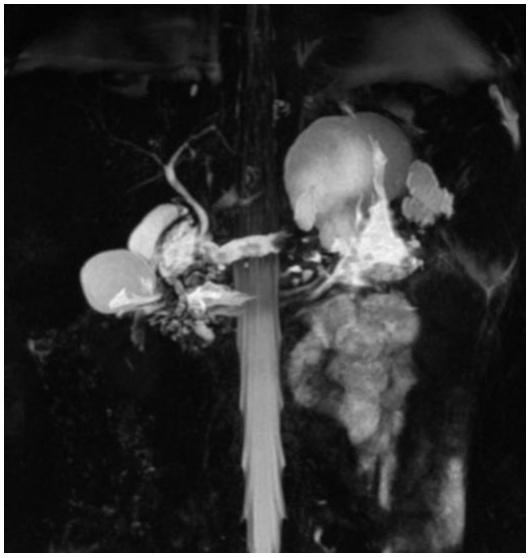

Figure 3. Magnetic Resonance Cholangiopancreatography showing multiple pancreatic stones and pseudocyst.

Preliminary management included the administration of low-fat soft diet, crystalloid fluid therapy, continuous drip/24-hour non-steroidal anti-inflammatory drug

(NSAID) analgesics, proton pump inhibitors (PPIs), and gastrointestinal probiotics. The patient was consulted to digestive surgeon and underwent Puestow Procedure (longitudinal pancreaticojejunostomy) with pre-operative antibiotics. (**Figure 4**)

Figure 4. Removed pancreatic stones (post-operative)

Postoperatively, the patient was admitted to the pediatric intensive care unit with nasogastric tube installed and received total parenteral nutrition for 24 hours, intravenous antibiotics, and epidural opioid analgesics with continuous analgesics drip. The second postoperative day, pancreatic functions were re-examined and pancreatic enzymes improved significantly (amylase 79 u/L and lipase 16 u/L). Diet was given gradually according to tolerance, starting from clear fluid to a soft diet-porridge with chopped side dishes. Gradual tapering down of analgesics was implemented. On the 8th postoperative day, the patient was discharged with no complaints of pain and clean surgical wound.

Discussion

Chronic pancreatitis is a rare disease in pediatric population, let alone causing the formation of pancreatic stones. The cause of pancreatitis in children is multifactorial, in which genetic mutations play the biggest role in cases of chronic pancreatitis. Four significant genetic mutations were found, namely CFTR, SPINKI, PRSS1, and CTRC with mutations often found in the PRSS1 gene.[6] In our case, genetic testing could not be performed due to cost constraints, thus, genetic risk factors could not be identified with certainty. In addition, other factors may also play a role, including disorders of the pancreatobiliary system (obstructive) such as pancreas divisum, gallstones, hypertriglyceridemia, drugs, alcohol, smoking, abdominal trauma, and autoimmune. In our patient, these risk factors were not observed.

Existing risk factors can stimulate excessive secretion of the exocrine pancreas cells which causes reflux of pancreatic enzymes and inflammation. The presence of gallstones can also obstruct the secretion of pancreatic enzymes, resulting in the

accumulation of zymogen that contains proenzymes such as trypsinogen in an inactive form. This accumulation causes the activation of proenzyme trypsinogen into an active form, namely trypsin in pancreatic cells, resulting in cell damage.[7]

PRSS1 gene mutations are associated with susceptibility to trypsinogen activation and continuous trypsin activity in the pancreas. Premature activation of trypsinogen to trypsin initiates a cascade of reactions leading to conversion of the proenzyme to the active enzyme. This results in auto-inflammation of the pancreas. PRSS1 gene mutations are most often associated with chronic pancreatitis because children with these mutations have a more rapid progression of chronic pancreatitis.[8]

The ongoing inflammatory process of the pancreas eventually leads to chronic pancreatitis. Pancreatic stone protein (PSP) plays an important role in the formation of pancreatic stones. Various factors such as genetic disorders cause a decrease in the PSP resulting in an increase in calcium carbonate in the pancreatic enzymes. The excessive accumulation of calcium carbonate eventually forms pancreatolithiasis.[5]

To diagnose pancreatitis in children, at least 2 of the following 3 criteria must be met: presence of abdominal pain, increased serum amylase and/or pancreatic lipase 3x above normal values and characteristic findings of pancreatitis based on radiological examination.[9,10] Abdominal pain is mainly felt in the epigastric region, both persistent and episodic, and may radiate to the back/left back waist. Pain can also be diffuse (spreading). The diagnosis of chronic pancreatitis is established when abdominal pain is consistent with the location of the pancreas and radiological findings suggest chronic pancreatic damage, evidence of exocrine and endocrine insufficiency of the pancreas, or biopsy results show histopathological abnormalities consistent with chronic pancreatitis.[4] In our patient, the main symptom was abdominal pain located in the pit of the stomach and left upper abdomen without any radiating pain. Abdominal pain was chronic because it was intermittent over the last 9 months, and with a severe pain scale (VAS) 8. Physical examination revealed epigastric and left hypochondria tenderness, in accordance with the anatomical location of the pancreas. Other symptoms that are often encountered include nausea and vomiting, abdominal distension, and irritability (restlessness). Jaundice can also occur if there is biliary obstruction. In our patient no jaundice was found.

Laboratory tests to diagnose pancreatitis were elevated serum lipase and pancreatic amylase at least 3x of the normal value.[9,10] In our patient, both pancreatic enzymes were found to be elevated 6x and >3x the normal value for amylase and lipase, respectively. The radiological modality of choice was transabdominal ultrasound because it was easy to perform, readily available, and without radiation. Chronic pancreatitis is mostly associated with obstruction, so MRCP may be the first choice

for evaluating the bile ducts and pancreas. With MRCP, ductal dilatation may be seen with or without strictures, stones (calculi), and pseudocysts.[11] In our patient, pancreatic ductal dilatation with stones and pseudocysts was observed which supported the diagnosis of pancreatic stones as a complication of chronic pancreatitis.

Treatment of pancreatitis needs to be managed comprehensively. Fluid resuscitation to maintain hydration status and prevent hypovolemia, adequate pain management, early enteral nutrition, are the three main pillars. Administration of 1.5-2x maintenance fluids is the first step by using crystalloids or crystalloid fluids containing dextrose as an alternative option.[9,10] In our patient, crystalloid solution was given.

Pain control is also a concern in managing pancreatitis. The administration of analgesics should follow the WHO "stepladder" of pain management. Non-opioid analgesics such as paracetamol and non-steroidal anti-inflammatory drugs (NSAIDs) are the first choice.[12] If long-term NSAIDs are to be used, patients need to be given histamine blocker-2 or proton pump inhibitor (PPI) to reduce gastrointestinal side effects. If the pain is still not resolved, opioid analgesics can be administered. Analgesics can be given orally or intravenously depending on tolerance. In our patient, NSAID analgesics together with PPI to reduce the side effects of gastric irritation. Intravenous administration was chosen because the patient showed symptoms of nausea and vomiting.

Oral nutrition can be done as early as possible without having to wait for the amylase and lipase values to improve. When possible and well tolerated, oral or enteral nutrition is preferred over total parenteral nutrition (TPN).[13] In our patient, oral nutrition was given despite amylase-lipase later improvement as in accordance with pancreatitis management and adjusting to patient tolerance.

Prophylactic antibiotics are not recommended as a routine therapeutic regimen, unless systemic signs, infection, or stones are found. In our case, antibiotics were given for the patient due to pre-operative prevention of infection.

Pancreatic stones (pancreatolithiasis) are late complication of chronic pancreatitis. Obstruction caused by these stones can cause unbearable severe pain. In the literature, it is stated that the Puestow procedure as an operative therapy for chronic pancreatitis produces favorable outcomes.[14] With regard to pancreatic stones, surgery is preferable rather than lithotripsy/endotherapy because it provides better long-term results, especially when multiple pancreatic stones are found.[6] Total pancreatectomy with islet auto transplant (TPIAT) may be considered in children with chronic pancreatitis who are still in pain and do not improve with other surgical methods.[15] In our case, surgery was chosen because multiple pancreatic stones were found.

Conclusion

Pancreatitis is often overlooked as one of the causes of abdominal pain in children. The incidence of chronic pancreatitis has increased in recent years. Chronic pancreatitis is a progressive inflammation that continues and cause irreversible damage to the structure and function of the pancreas, in which can also be accompanied by pseudocysts. Pancreatic stones are one of the late complications of chronic pancreatitis. Surgery is an alternative therapy in cases of pancreatitis accompanied by stones. A more careful diagnostic approach and a greater degree of suspicion of pancreatitis as one of the differential diagnoses are needed for recurrent abdominal pain in children. With the right diagnosis, adequate management can be carried out to prevent further complications of pancreatitis.

Conflict of Interest

None declared.

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