Intrahepatic Pancreatic Pseudocyst: Case Series

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ABSTRACT

Intrahepatic pseudocyst is a very rare complication of pancreatitis. Lack of experience and literature makes diagnosis and management of intrahepatic pseudocyst very difficult. Majority of published cases were managed by either percutaneous or surgical drainage. Less than 30 cases of intrahepatic pseudocysts have been reported in the literature and there is not a single report of endoscopic ultrasound guided management of intrahepatic pseudocysts. Here we report a case series of 2 patients who presented with intrahepatic pseudocysts and out of which first case was successfully managed by EUS guided drainage. Our second case is also the youngest patient presented with intrahepatic pseudocyst till now.

INTRODUCTION

A pancreatic pseudocyst is a collection of pancreatic fluid located in or around the pancreas. Pancreatic pseudocysts are encased by a non-epithelial lining of fibrous, necrotic and granulation tissue secondary to pancreatic injury. They develop at least four weeks after this damage has occurred. Pseudocyst is a complication of acute or chronic pancreatitis which can occur at any site in the abdomen and even in the mediastinum, but the intrahepatic location of pancreatic pseudocyst is a very uncommon event [1, 2]. Only about 28 cases have been reported in the literature and there is not a single report of endoscopic ultrasound (EUS) guided management of intrahepatic pseudocysts. Here we report a case series of 2 patients who presented with intrahepatic pseudocysts and out of which first case was managed by EUS guided drainage.

CASE SERIES

Case #1

A forty-five-year-old male presented with intermittent epigastric abdominal pain since 1 year which was boring in character, radiating to back. Patient had increasing severity of pain since last 2 months which was now continuous. He also developed intermittent episodes of vomiting which were bilious, nonprojectile, without blood and unrelated to food. He also developed generalized abdominal distention since last 1 month. However he did not have significant weight loss, gastrointestinal bleeding, pedal edema, jaundice, fever. His past medical history and family history was not significant. He was chronic alcoholic since last 15 years with intake of approximately 90 gram per day. On physical examination he had ascites and left lobe of liver was palpable 3 cm below xiphisternum. His blood investigation showed Hb-9.8 gm%, platelet-215000/ mm³, leucocyte count-4300/mm³, total bilirubin 0.6 mg% (0.2-1.2 mg %), ALT-12 IU/L (0-40IU/L), AST-22 IU/L (0-40IU/L) with Serum alkaline phosphatase-455 (upper limit of normal -306 IU), serum creatinine 0.9 mg/dL (0.7-1.2 mg/dL). His serum amylase level was 379 IU (reference range: 30-80 IU/L). Serum lipase level was 789 IU (reference range: 45-180 IU/L). His HIV ELISA was negative. Chest roentgenogram was normal. CT scan of the abdomen with pancreas protocol (Figure 1) showed atrophic pancreas with necrosis in head and neck with intrapancreatic and extrapancreatic collection suggestive of walled off collections. Pancreatic duct was measuring 7 mm. There was large well defined collection was seen in subcapsular region of left lobe measuring 14.5×9.2×13 cm which was not communicating with pancreatic duct. Moderate ascites was present. There was complete thrombosis of main, right and left branch of portal vein with cavernoma formation. Spleen was normal in size. MRCP (Magnetic resonance cholangiopancreatography) was suggestive of same finding as CT scan of abdomen with no communication of intrahepatic pseudocyst with pancreatic duct. Ascitic fluid SAAG (serum ascites albumin gradient) ratio was 0.9. Ascitic fluid amylase level was 9600 IU/L. Ultrasound guided fine needle aspiration of hepatic cyst fluid showed clear fluid with an amylase level of 12,300 IU/L (reference range: 0-160 IU/L) and a total bilirubin level of 0.6 mg/dL (reference range: 0.2-1.0 mg/dL). Upper gastrointestinal endoscopy showed no esophageal or gastric varices or portal hypertensive gastropathy. Patient was started on nasojejunal feeding

Received January 24th, 2016 - Accepted March 30th, 2016 **Keywords** Endosonography; Pancreatic Pseudocyst **Abbreviations** EUS Endoscopic Ultrasound **Correspondence** Dhaval Gupta Room no.2, First Floor, College building Department of Gastroenterology Lokmanya Tilak Municipal Medical College &Hospital (LTMMC & LTMGH) Sion, Mumbai 400022, Maharashtra, India **Phone** +919819277795 **Fax** +9102224076100 **E-mail** drdhavalgupta@gmail.com

but was stopped as patient could not tolerate feeding. We performed EUS guided transgastric cystogastrostomy by 180 series Olympus linear echoendoscope. Pre procedure antibiotic was given. We put 10 french double pigtail stent **(Figure 2)** and another 10 french nasocystic drainage tube. Patient had a gradual subjective improvement and the abdominal pain subsided within few days. Serial ultrasonograms revealed a progressive decrease in the size of intrahepatic pseudocyst. After 6 weeks of nasocystic drainage placement, complete resolution of pseudocyst was seen and pigtail was removed endoscopically.

Case #2

Ten-year-old male child presented with complain of right hypochondriac and epigastric abdominal pain since 4 months. Pain was dull aching, continuous nonradiating with no relation with food intake. He had complaint of weight loss of 3 kg in 5 months. Otherwise he had no complaint of vomiting, abdominal distention, gastrointestinal bleeding, jaundice, fever, growth retardation. Patient had past history of severe epigastric abdominal pain which was continuous, boring in character, radiating to back suggestive of acute pancreatitis. He also had previous history of blunt abdominal trauma before 2 years. There was no significant family history. On physical examination, liver was palpable 2 cm below costal margin. His blood investigations were normal including complete hemogram, ranal function and liver function test. His serum amylase level was 179 IU (reference range: 30- 80 IU/L). Ultrasound of abdomen showed large cystic lesion in liver. CT scan of abdomen with pancreas protocol (Figure 3) showed multiple hypodense lesion suggestive of intrapancreatic pseudocysts. There was large intrahepatic cyst occupying majority of liver parenchyma. Ultrasound guided aspiration of hepatic cyst fluid showed clear fluid with an amylase level of 5400 IU/L (reference range: 0-160 IU/L) and a total bilirubin level of 0.4 mg/dL (reference range: 0.2-1.0 mg/ dL). We also did MRCP but there was no pancreatic ductal leakage and intrahepatic pseudocyst was not communicating with pancreatic duct and did not show changes of chronic pancreatitis. Patient was started on nasojejunal feeding but was stopped as patient was not tolerating feeding. We did radiology guided percutaneous drainage with placement of 8F pigtail catheter. Prior antibiotic was given. Subsequently patient relieved from abdominal pain. Initial fluid output was 400 milliliter on day 1 which subsequently reduced



Figure 1. CECT scan of abdomen showing large well defined intrahepatic pseudocyst in subcapsular region of left left lobe of the liver with moderate ascites. (asterix in both figures)



Figure 2. EUS guided transgastric intrahepatic cystogastrostomy. (a). showing large intrahepatic pseudocyst and (b). showing luminal end of the pigtail stent.



Figure 3. CECT scan of abdomen showing large intrahepatic pseudocyst in the subcapsular region in the right lobe of the liver occupying majority of liver parenchyma (asterix in both figures) with multiple intrapancreatic pseudocysts.

upto nil output at the end of 2 weeks. Follow up ultrasound of abdomen showed gradual shrinkage of size of pseudocyst and complete resolution after 2 weeks of pigtail insertion. Pigtail was removed after 2 weeks. After 8 weeks of follow up patient was asymptomatic without any ultrasonographic evidence of pseudocyst.

DISCUSSION

Pancreatic pseudocysts have been well recognized and common complication of both acute and chronic pancreatitis. However, intrahepatic location of pseudocyst have rarely been identified [1, 2]. Only about less than 30 cases have been reported in the literature till now and all were managed with either percutaneous, endoscopic transpapillary or surgical drainage procedures. Commonly, intrahepatic pseudocysts are single and involve left lobe of liver. However, multiple and right lobe intra-hepatic pseudocysts have also been described [2, 3, 4]. The pathophysiology of intrahepatic pseudocysts can be explained by two mechanisms [3, 5]. The first proposed mechanism is leakage of the pancreatic juice in pre-renal space and thereafter eroding through the posterior layer of the parietal peritoneum and into the lesser sac. Lesser sac collection then follow the path along the lesser omentum or gastrohepatic ligament toward the liver leading to the formation of left lobe subcapsular collections. This may be the mechanism in our first case. Second theory is tracking of pancreatic juice along the hepatoduodenal ligament from the head of pancreas to the porta hepatis resulting in formation of intraparenchymal collections. This may be the mechanism of intrahepatic pseudocysts in our second case. Pseudocysts which forms as per the first mechanism are mainly subcapsular in location and are biconvex in shape. Intraparenchymal pseudocysts formed as a result of the second theory are located away from the liver capsule and are located near branches of porta hepatis [6]. Intrahepatic pseudocyst is a diagnostic challenge because it is not usually considered in the differential diagnosis of cystic hepatic lesions. Amylase-rich fluid on aspiration and communication of pseudocyst with disrupted pancreatic duct on imaging are indicative of diagnosis. However, none of our case had communication with pancreatic duct.

There is no consensus regarding the management of intrahepatic pseudocysts. Majority of the published intrahepatic pseudocysts were managed by either Surgical, radiologically guided percutaneous drainage/aspiration or by ERCP guided transpapillary drainage [7, 8, 9, 10]. There is not a single case report of EUS guided management of intrahepatic pseudocyst. We managed our first case of intra-hepatic pseudocysts successfully by EUS guided transgastric cystogastrostomy alone. As in our first case there was no communication with pancreatic duct, ERCP guided transpapillary drainage would not be beneficial. Percutaneous drainage is associated with risk of incomplete drainage, infection, catheter site fistula formation. Similarly sugery is also associated with its own risk. EUS guided drainage is a relatively safe procedure in expert hand, provides intestinal drainage of fluid with minimal risk of bleeding as it avoids collaterals in between. There is no study which has compared the efficacy of percutaneous, endoscopic or surgical drainage. Nasocystic drainage has advantage that blocked nasocystic tube can be opened up by flushing and aspiration and therefore obviating the need of a repeat procedure. We managed our second case of intrahepatic pseudocyst with percutaneous drainage, as pediatric EUS endoscope was not available and cyst was also quite away to be managed by transgastric EUS. Our second patient is also the youngest patient presented with intrahepatic pseudocyst till now.

To conclude, an intra-hepatic pancreatic pseudocyst is a very rare complication of pancreatitis. Presence of liver lesion in presence of pancreatitis should raise possibility of intrahepatic pseudocyst. However, it can be managed by either percutaneous radiological, ERCP or surgical drainage, EUS guided drainage is also a safe and viable option when pseudocysts are accessible. And finally, intrahepatic pseudocyst can also present in pediatric age group.

Author's contribution

Dhaval Gupta worked up completely these cases. Meghraj Ingle and Nilesh Pandav assisted during work up of these cases. Nirav Pipalia and Kaivan Shah helped in collecting references. Prabha Sawant has supervised and critically evaluated these cases.

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Conflict of interest

There is no conflict of interest of any author.

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