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Case Report

Communicating pancreatic and splenic pseudocysts: A case report ☆

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ABSTRACT

A splenic pseudocyst is an encapsulated mature collection located within the splenic tissues. It is usually asymptomatic and is detected incidentally during either radiological workup or laparoscopy for other reasons. Our patient is a 66-year-old male presenting with a history of weight loss, early satiety, and constipation, found to have a splenic pseudocyst communicating with a preexisting pancreatic cyst. Cystic lesions of the spleen can be divided into primary and secondary types. Secondary splenic cysts (pseudocysts) are residues of either earlier infection, trauma, or infarction. Management approaches to splenic cysts are either conservative or surgical according to the symptoms and size of these cysts. Most splenic cysts are discovered incidentally either during radiological workup or laparoscopy. Such entities require the combined effort of surgeons, gastroenterologists, and radiologists to provide the maximum care for these patients.

Introduction

A splenic pseudocyst is an extremely rare condition with an incidence of 0.07 in the general population [1]. A splenic pseudocyst is an encapsulated mature collection located within the splenic tissues. Most splenic pseudocysts are asymptomatic and discovered incidentally during imaging studies for other indications [2,3]. Blunt abdominal trauma is the most common etiology, representing 75% of cases [4]. Infarction, infection (tuberculosis, mononucleosis, or malaria), and degenerative diseases are other possible etiologies for these kinds of splenic cysts [3,5]. If the cyst is asymptomatic with a diameter of less than 5 cm, conservative management may be useful whereas surgical management is preferred for symptomatic cysts or if the cyst is larger than 5 cm [6]. We present an in-

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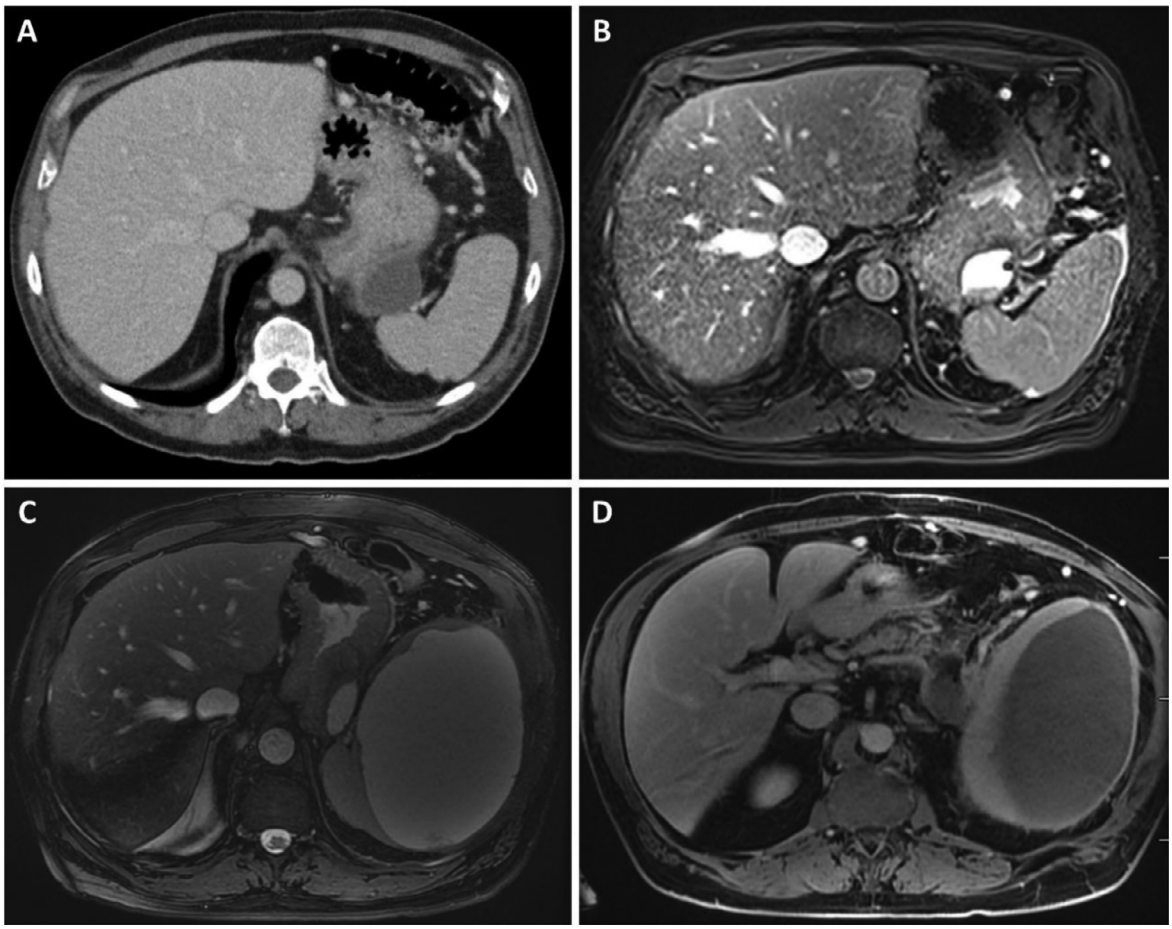

Fig. 1 – Axial CT scan (A) showing an irregular loculated fluid collection in the tail of the pancreas area measuring 1.6 x 5.5 x 4.5 cm, axial T2 fat sat-weighted MRI image (B) showing persistent complex cystic mass in the pancreatic tail with some heterogeneous signal intensity. Axial 2D FIESTA and T1 fat-suppressed, postcontrast-weighted MRI (C and D) showing mild interval decrease in size of the 2 communicating cystic lesions at the pancreatic tail with interval development of a huge subcapsular fluid collection along the lateral aspect of the spleen. Within these fluid collections there are no enhancing soft tissue components.

teresting case of a huge splenic cyst communicating with a preexisting bilobed pancreatic tail cyst in a 66-year-old male patient.

Case presentation

A 66-year-old male presented to the outpatient clinic with a history of weight loss, early satiety, and constipation. Upon review, there was no history of chronic pancreatitis or alcohol consumption. Physical examination was unremarkable. A computerized tomography (CT) scan was ordered and showed an irregular loculated fluid collection in the tail of the pancreas measuring 1.6 × 5.5 × 4.5 cm (Fig. 1A). The serum amylase was elevated (123 U/L). Given the previous finding on the CT scan, the patient was referred for evaluation by endoscopic ultrasound-fine needle aspiration (EUS-FNA). Aspiration of a total of 30 mL showed turbid brown cystic content with carcinoembryonic antigen (CEA) of 2 ng/mL (normal range is 0-2.5 ng/mL), and amylase >6000 U/L (normal range is 40-140 U/L). Based on the fact that the source of pancreatitis and ongoing serologic elevations in lipase was unclear, surveillance imaging to assess for the persistence of the lesion was planned.

Six months later, magnetic resonance cholangiopancreatography (MRCP) (Fig. 1B) was performed and showed a persistent complex cystic mass in the pancreatic tail with some heterogeneous signal intensity. The lesion was bilobed in appearance and grossly measured 4.1 × 4.2 × 8.2 cm in craniocaudal dimension. Peripheral wall enhancement was noticed postcontrast, which raised the suspicion of a possible neoplastic process. A follow-up-enhanced MRCP in 3 months to reassess the progression of the lesion was planned given that the size of the lesion had not significantly changed, the very distal location of the lesion (so the majority of pancreatic parenchyma appears unaffected), and the absence of truly worrisome new enhancement on the MRCP that would be significant for malignancy.

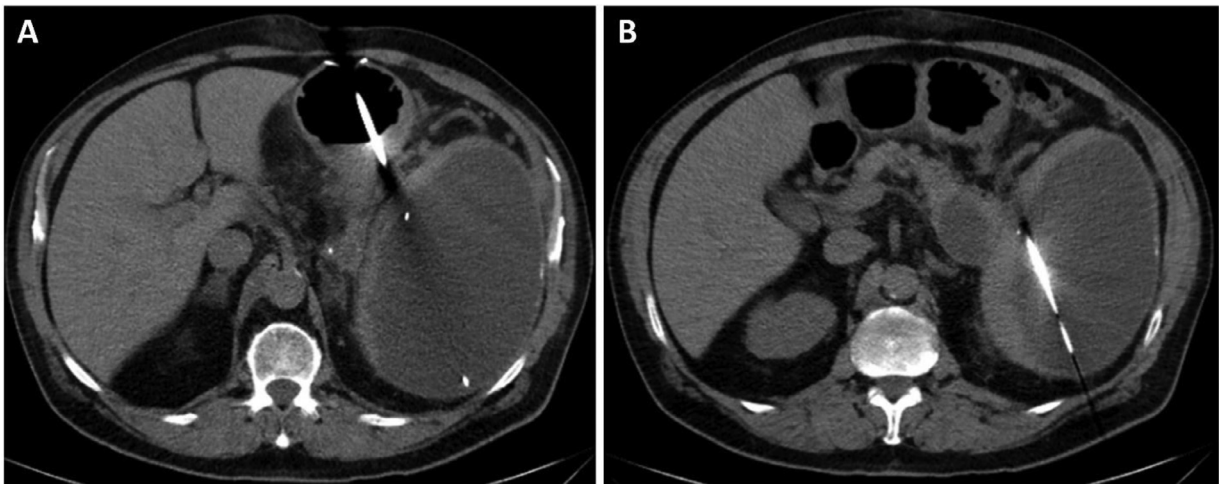

Fig. 2 – Axial CT scan (A and B) demonstrate successful placement of a 14 French transgastric cystogastrostomy tube with distal pigtail loops within the splenic subcapsular space.

The follow-up MRCP (Fig. 1C and D) showed a mild interval decrease in the size of the 2 communicating cystic lesions at the pancreatic tail with interval development of a huge subcapsular fluid collection along the lateral aspect of the spleen, which appeared also to communicate with these 2 previously seen pancreatic tail collections. The splenic parenchyma was stretched along the anterior lateral aspect of this subcapsular fluid collection with normal splenic parenchymal enhancement. Within these fluid collections, there were no enhancing soft tissue components.

Due to the significant increase in the size of the pancreatic pseudocyst, which now involved the spleen, a percutaneous CT-guided 14 French cystogastrostomy tube with distal pigtail loops was successfully placed within the splenic subcapsular space (Fig. 2A and B). A suitable access site was identified by CT scan. Two cope GI T-fasteners were placed into the gastric lumen and their location was confirmed using a CT scan. The gastric lumen was traversed with an 18-gauge Chiba needle with the tip of the needle accessing the splenic subcapsular space via a transgastric approach under CT scan guidance. A 0.035 soft-tipped Amplatz guidewire was placed through the transgastric access and into the splenic subcapsular space. Under direct fluoroscopic visualization, a modified 14 French Cook multipurpose drain in which 3 extra side holes were cut and positioned so that these side holes resided within the gastric lumen, was placed over the wire (Fig. 3). The drain was advanced through the anterior and posterior wall of the stomach over the wire and the distal loop was coiled within the splenic subcapsular space. About 1200 mL of murky brown fluid was aspirated from the distal pigtail loop within the subcapsular collection. A sample of this fluid was sent to the laboratory for analysis. The drain was successfully removed 2 days later but the Intuit G tube was left in place. Serial imaging was performed after the drainage which showed that there was a mild interval decrease in the size of the fluid collections.

Due to significant residual fluid, a surgical management approach was warranted. Given the patient's history of necrotizing pancreatitis and cholelithiasis, an open cholecystectomy was performed, followed by a gastrocutaneous fistula takedown. Pancreatectomy and splenectomy were then performed. A firm mass in the left upper quadrant consistent with a fluid collection around the spleen was found. An intraoperative ultrasound was performed to ensure that the pancreatic and splenic vascular anatomy was identified appropriately. The surgeon elected to proceed by incising the peritoneum overlying the pancreas at its inferior border and dissecting the inferior border free and then dissecting the superior border. There was a significant amount of saponified and calcified fat which was densely adherent to the surrounding tissues. During the course of dissection, a significant amount of pancreatic fluid was drained from the pseudocyst and was sent for culture. The distal pancreas and the spleen were removed and sent to pathology. There were no acute complications apparent. The patient was extubated and transported to post-anesthesia care unit in stable condition. The histopathology of the specimens came back as fibrotic cyst wall of the distal pancreas and spleen which was compatible with a pancreatic pseudocyst, the pancreas had mild peri-ductular fibrosis, suggestive of mild chronic pancreatitis, and the spleen had no significant histologic abnormality. The patient's symptoms improved after the surgery, and no chronic complications were observed on follow-up 1 month after the surgery.

Discussion

Cystic lesions of the spleen are entities of uncommon occurrence. Only about 800 cases of splenic cysts have been documented worldwide [7]. In 1978, F G Robbins evaluated 42,327 autopsies across 25 years period and reported that 32 subjects (0.75 in 1000) exhibited benign cysts of the spleen as an incidental finding at postmortem. Since Andral described the first instance of a splenic cyst in 1829, the categorization of these lesions has developed and changed over the years [8].

Martine divides cystic lesions of the spleen into primary and secondary types, determined by the presence of an epithelial lining which is the currently followed classification [9]. The presence of an epithelial lining distinguishes primary (true) cysts. On the other hand, the absence of an epithelial

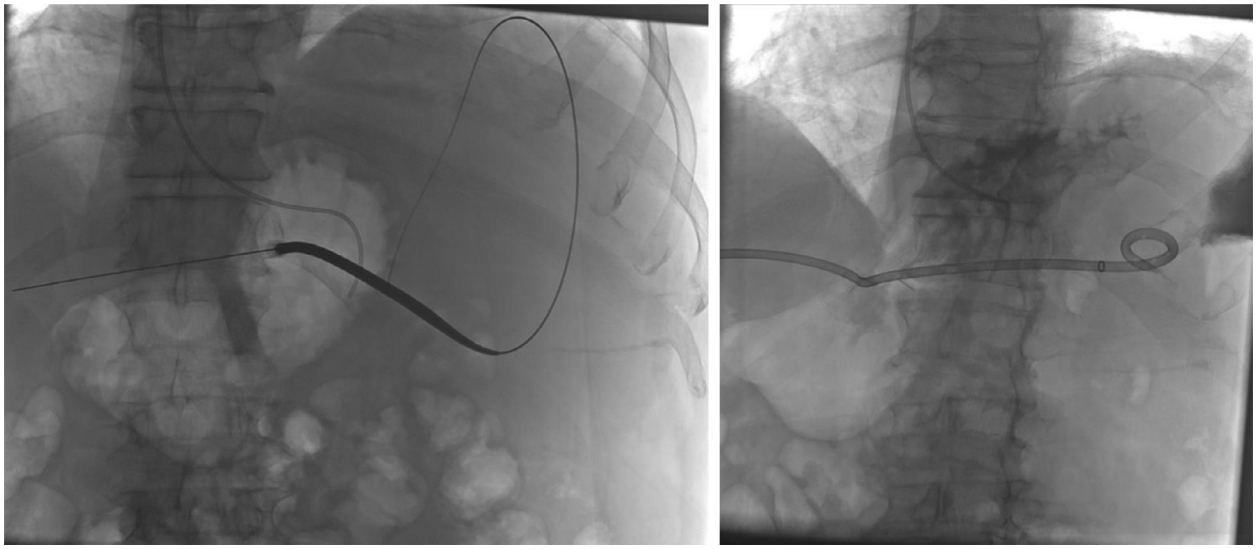

Fig. 3 – Placement of modified 14 French Cook multipurpose drain over the wire, under direct fluoroscopic visualization.

lining distinguishes secondary (false or pseudo-) cysts, which also lack a capsule [9,10].

Primary splenic cysts comprise parasitic as well as non-parasitic etiologies. Primary parasitic splenic cysts are the most common cause of splenic cysts worldwide. They are typically found in endemic regions and are triggered mostly by Echinococcus granulosus infection [11–13]. Nevertheless, primary nonparasitic splenic cysts can be further subdivided into congenital and neoplastic [2]. Approximately 80% of primary nonparasitic cysts are seen in individuals under the age of 20, indicating a congenital nature [14].

However, secondary splenic cysts (pseudocysts) are the sequela of either earlier infection, trauma, or infarction [3,15,16]. Splenic damage leading to secondary cysts is most prevalent in abdominal trauma [17]. In fact, the most common cause of secondary splenic cysts is trauma, which is estimated to be associated with approximately 75% of secondary cases [18]. As a result of the trauma, a hematoma is formed which will eventually be resorbed forming a residual cystic collection. On the other hand, nontraumatic etiologies are related to either infection or infarction [2]. Regarding infections, pseudocysts are frequently related to tuberculosis, mononucleosis, or malaria [19–21]. Whereas in infarction, an embolus to the spleen can lead to cyst development by activating hydrolytic enzymes, which causes liquefactive necrosis [6].

Vascular cysts of the spleen, which are called peliosis, is a rare condition consisting of several irregular blood-filled cavities. This condition is typically discovered at autopsies. This has been linked to a number of underlying severe conditions, including tuberculosis, acquired immunodeficiency syndrome, and malignancies, in addition to prolonged alcohol intoxication, steroids, and others [22,23].

A significant percentage of splenic cysts are asymptomatic [24]. Most splenic cysts are discovered incidentally either during radiological workup or laparoscopy, or if the patient develops symptoms, which are related to the size of the cyst and the development of complications such as hemorrhage, infection, or rupture [25]. The most prevalent complaint is a dull pain in the left upper abdominal region and left shoulder [2]. Moreover, nausea, vomiting, early fullness, and respiratory problems are all common symptoms [3].

Indeed, a radiological workup is essential in the diagnosis, the selection of the most appropriate operational strategy, and in the following up of patients in both conservative management and post-operatively [2]. Either primary or secondary cysts can have calcium deposits or trabeculations in their walls, as well as peripheral septations and remnant materials. As a result, the differentiation between primary and secondary splenic cysts is challenging radiologically [26].

In children, ultrasonography (US) is the imaging modality of choice. On ultrasound, the typical splenic cyst appears as an anechoic, spherical, homogenous region with significant echo enhancement and a thin, smooth wall [27]. However, thin septations, uneven cyst surfaces, and a mixed distribution of echogenicity from intra-cystic debris or bleeding, as well as outer highly echogenic foci with distant shadowing due to cyst wall calcifications, can all contribute to a more complicated image [28].

Computerized tomography (CT) scan is also used, in which, splenic cysts appear typically as a well-defined, near water density, spherical structure with a thin or undetectable wall with no rim enhancement. Septations and calcifications of the cyst wall are clearly visible using CT scan [25,28].

By using magnetic resonance imaging (MRI), splenic cysts often exhibit a signal intensity equivalent to that of water on both TI- and T2-weighted MR imaging. Based on the content of the cystic fluid, the signal intensity on TI-weighted imaging may be higher than expected, although the signal intensity on T2-weighted images stays high. MRI can also be used to gain a better understanding of the interaction between the cyst, the spleen, and the surrounding structures [28,29].

In order to determine if the lesion is pulsatile or not, Doppler US is beneficial [28,30]. Moreover, due to the widely varied structure of the splenic arteries, angiography is often necessary for preoperative assessment if a partial splenectomy is contemplated [31].

In addition, if a splenic cyst is thought to be benign, chiefly based on diagnostic imaging workup, percutaneous US-assisted cyst puncture has shown its effectiveness not only for establishing the diagnosis (fluid analysis for microscopic organisms, enzymes, etc.) but also for reducing the size of the cyst [32,33]. However, fluid analysis cannot make a distinction between an actual cyst and a pseudocyst, and the only approach to determine the type and etiology of a splenic cyst is by histopathological investigation [34,35]. Nevertheless, the potential possibility of introducing cancerous cells into the peritoneal cavity or along the needle route should be considered, despite the fact that this risk is insignificant [28].

Management approaches to splenic cysts are either conservative or surgical. Conservative management is applied if the cyst is asymptomatic and measures less than 5 cm. On the other hand, surgical management is reserved for symptomatic cysts or if the cyst is larger than 5 cm [6,25].

Since splenic cysts are considered rare, treatment was formerly based on personal preference. Total splenectomy was the management of choice but because of the realization of the spleen's critical activities as well as the risks of intense post-splenectomy infection, conservative splenic approaches are the preferred management strategies [25]. These include laparoscopic marsupialization or fenestration for cysts that are positioned on the surface, and partial splenectomy in the case of cysts found deep within the spleen [36–39]. For example, in polycystic conditions when the cysts are unreachable for fenestration or marsupialization, also in case of a huge cyst that is almost entirely covered by splenic tissue as there is a chance of intractable splenic hemorrhage, or in case of the development of intractable bleeding during the surgery a total splenectomy may be required [25,40].

Conclusion

Since splenic cysts are considered rare, treatment was formerly based on personal preference. Conservative approaches including cyst aspiration, laparoscopic marsupialization or fenestration, or partial splenectomy are the preferred management strategies over total splenectomy. Such an approach requires comprehensive care from surgeons, gastroenterologists, and interventional radiologists.

Ethical approval

Ethical approval for case reports and case series are waived according to the ongoing regulations of University of Michigan.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. Written consent is available for review by the Editor-in Chief of this journal on request.

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