

CASE REPORT

A Wayward Cyst

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ABSTRACT

Context Pseudocysts are a common complication of acute and chronic pancreatitis. These are usually located within the pancreas but they can occur at other sites as well, including the mediastinum, neck, pelvis and rarely in the liver as in our case. The diagnosis of intrahepatic pancreatic pseudocyst relies on the demonstration of a high amylase level in the sampled cystic fluid in the absence of infection or neoplasm. **Case report** A 60-year-old man with a history of chronic pancreatitis presents with a clinical and laboratory picture suggestive of acute exacerbation of his pancreatitis. A computed tomogram (CT) scan of the abdomen revealed a pancreatic pseudocyst and a cystic lesion involving both lobes of the liver. CT diagnostic aspiration of the intrahepatic cyst revealed high amylase level (greater than 20,000 U/L). The cyst was treated with percutaneous drainage with complete resolution of the cyst. **Conclusion** In the setting of pancreatitis, intrahepatic pancreatic pseudocyst should be considered in the differential diagnosis of cystic lesion of the liver.

INTRODUCTION

The differential diagnosis of cystic lesions of the liver is broad and includes pyogenic liver abscess, echinococcal cyst, amebic abscess, biloma, malignancy and pseudocysts among others. Pseudocysts are most commonly seen in the pancreas as complications of acute and chronic pancreatitis, though on rare occasions pseudocysts can be found outside the pancreas. We describe such a case of a pseudocyst involving both lobes of the liver and discuss the diagnosis, management and review of the literature of this interesting entity.

CASE REPORT

A 60-year-old man with a history of chronic pancreatitis secondary to chronic alcohol abuse was admitted following two days of epigastric pain, nausea, and vomiting. There was no history of fever, jaundice, hepatitis, trauma or weight loss. His medications included aspirin, captopril, alendronate, metoprolol and omeprazole. On physical examination he was afebrile with normal vital signs. His abdominal examination

was significant for mild tenderness in the epigastrium and hepatomegaly. Laboratory testing showed hemoglobin of 13.9 g/dL (reference range: 13.6-17.3 g/dL), white blood cell count 16,000 cells/mm³ (reference range: 4,800-10,500 cells/mm³), serum amylase 656 U/L (reference range: 30-111 U/L), serum lipase 4,590 U/L (reference range: 46-218 U/L), bilirubin 0.9 mg/dL (reference range: 0.2-1 mg/dL), aspartate aminotransferase 281 U/L (reference range: 7-56 U/L), alanine aminotransferase 89 U/L (reference range: 15-46 U/L), and alkaline phosphatase 135 U/L (reference range: 46-218 U/L).

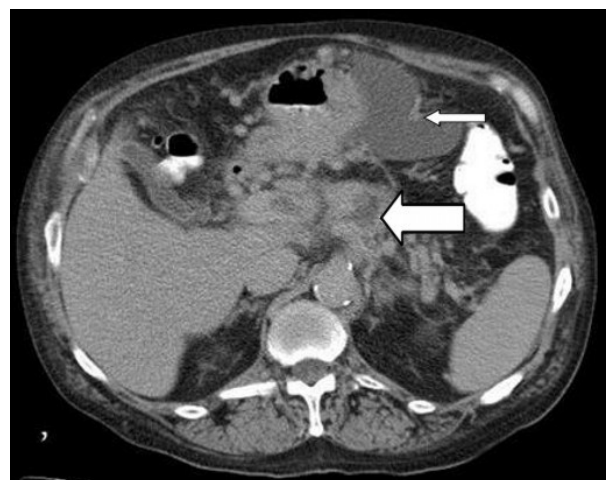


Figure 1. Computed tomography scan of the abdomen shows a small pancreatic pseudocyst (wide arrow) and fluid collection around the stomach (narrow arrow).

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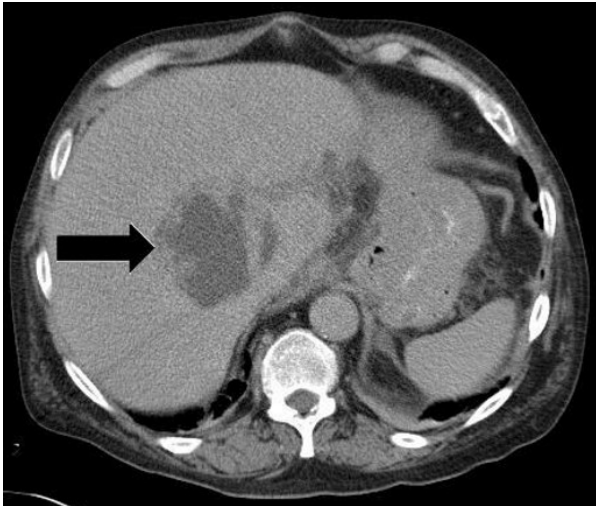


Figure 2. Computed tomography scan of the abdomen shows a large (8x8x7 cm) cystic lesion in the liver (arrow).

A computed tomogram (CT) scan of the abdomen revealed a small pancreatic pseudocyst (Figure 1) as well as a large (8x8x7 cm) cystic lesion involving both lobes of the liver which was read as suspicious for an abscess or necrotic neoplasm (Figure 2). CT guided diagnostic aspiration of the cyst drained 95 mL of green colored fluid which showed no organisms on Gram stain and was sterile on bacterial and fungal cultures. Cytological examination of the fluid did not reveal any malignant cells. The amylase level in this fluid was greater than 20,000 U/L which confirmed the diagnosis of an intrahepatic pseudocyst. At the time of CT guided aspiration, the pseudocyst was treated with percutaneous drainage with the placement of an 8F pigtail catheter. The pigtail catheter was removed 8 days later. There was both clinical improvement and near total resolution of the pseudocyst—11 days after removal of the pigtail catheter (Figure 3). A repeat CT scan one year later showed complete resolution of the cyst (Figure 4).



Figure 3. Computed tomography scan of the abdomen shows near total resolution of the cyst (arrow).

DISCUSSION

Pancreatic pseudocysts are a well recognized and common complication of acute and chronic pancreatitis and are also seen following trauma [1]. These pseudocysts are fluid filled cavities surrounded by fibrous or inflammatory tissue without epithelial coverage. The incidence of pseudocysts is 5-16% following acute pancreatitis and 20-40% in chronic pancreatitis [2]. Pancreatic pseudocysts most commonly occur within the body or the tail of the pancreas; however, they can form at various sites outside the pancreas, including the mediastinum [3], neck [4], pelvis [5] and rarely in the liver as in our case.

Most of the described cases of intrahepatic pancreatic pseudocysts occurred in the left lobe of the liver, although occasionally they have been seen in the right lobe as well [6]. A literature review by Mofredj *et al.* in 2000 revealed 26 reported cases of intrahepatic pancreatic pseudocysts that had been documented by sonography, CT or surgical exploration. The right lobe of the liver was involved in only three of these cases [7]. Since Mofredj's review, three more cases of intrahepatic pseudocyst formation have been reported [8, 9, 10]. We describe the rare occurrence of intrahepatic pseudocyst in both lobes of liver with only one such case previously reported [11].

It is believed that the location of pseudocyst formation depends on the location of the release of the pancreatic enzymes and hence the path of pancreatic digestion. It is hypothesized that if the pancreatic proteolytic enzymes leak and spread from the posterior part or tail of the pancreas to the lesser sac and then follow the path along the hepatogastric ligament, they may form a pseudocyst in the left lobe of liver. Similarly, if pancreatitis predominantly involves the pancreatic head and the enzymes exude and follow along the hepatoduodenal ligament to the porta hepatis then the pseudocyst may form in the left or right lobe of the liver.



Figure 4. Computed tomography scan of the abdomen done 1 year later shows complete resolution of the cyst.

There may be another explanation for the presence of a high level of amylase in these hepatic cysts. Both the liver and pancreas arise from the same region of the endoderm and transdifferentiation (term used to describe the conversion of one cell type to another) or plasticity (term means that a stem cell from one adult tissue can generate the differentiated cell types of another tissue) of liver into pancreas cells and *vice versa* has been described *in vitro* [12, 13]. In addition, hepatic bile cells are capable of secreting amylase [14]. We hypothesize that intrahepatic pancreatic pseudocyst formation may represent transdifferentiation of the liver cells into pancreatic cells with the production of amylase, rather than the previously described hypothesis of pancreatic enzymes extending into the liver. Chronic pancreatitis could create the environment suitable for transdifferentiation. However, this theory cannot explain the occurrence of pseudocysts at other more peripheral sites like the neck and the pelvis.

Clinically, patients with intrahepatic pancreatic pseudocysts can present with continuous epigastric pain or recurrence of pain after initial resolution of acute pancreatitis [7]. On physical examination there may be a palpable abdominal mass [6, 15, 16] or less frequently hepatomegaly [11, 17, 18] or jaundice [16]. Laboratory tests usually reveal elevation of the pancreatic enzymes but with normal liver enzymes [7]. The elevated liver enzymes in our case were most likely due to hepatocellular damage from enzymatic proteolysis as well as from a pressure effect of the pseudocyst on the liver parenchyma.

The diagnosis of an intrahepatic pancreatic pseudocyst is established with the demonstration of a high amylase level in the sampled cystic fluid in the absence of infection or neoplasm. An amylase level greater than 479 U/L has 73% sensitivity and 98% for diagnosing pancreatic pseudocyst [19]. CT scans of the abdomen typically show round or oval areas of decreased attenuation though occasionally pseudocysts may have areas of high attenuation if there is blood or necrotic debris involved.

Cystic lesions of the liver have a broad differential diagnosis including simple cysts, polycystic disease, hemangiomas, abscesses, hydatid cysts, necrotic neoplasms, Caroli's disease, and other rare causes. Great care should be taken to exclude malignancy as a possible etiology in these cases [20]. For pancreatic pseudocysts, endoscopic retrograde cholangiopancreatography (ERCP) is useful in identifying a communication between the pseudocyst and the pancreatic duct in 40-69% of cases and aids with the development of an operative strategy when surgery is needed [21]. For intrahepatic pancreatic pseudocysts, ERCP may be necessary to demonstrate a normal biliary tree and to exclude biliary obstruction or malignancies [22]. Endoscopic pancreatic duct drainage and stenting can also be effective for treating pancreatic pseudocysts [23], a method that still needs to be explored for treating intrahepatic pancreatic pseudocysts.

While most pseudocysts, pancreatic or extrapancreatic, resolve spontaneously and require no intervention, they can be complicated as in our case by persistent nausea, vomiting, and abdominal pain, or can rupture, form fistulas, obstruct the common bile duct or become infected. When so indicated, percutaneous drainage of the pseudocyst is the treatment of choice though surgery is occasionally required.

CONCLUSION

Our report reminds internists to include intrahepatic pancreatic pseudocyst in their differential diagnosis of a cystic lesion of the liver, especially in the setting of pancreatitis. Fine needle aspiration of the lesion and demonstration of a high amylase level can exclude other causes and confirm the diagnosis of an intrahepatic pancreatic pseudocyst.

Conflict of interest The authors have no potential conflict of interest

References

1. Baron TH. Treatment of pancreatic pseudocysts, pancreatic necrosis, and pancreatic duct leaks. *Gastrointest Endosc Clin N Am* 2007; 17:559-79. [PMID 17640583]
2. Aghdassi A, Mayerle J, Kraft M, Sielenkämper AW, Heidecke CD, Lerch MM. Diagnosis and treatment of pancreatic pseudocysts in chronic pancreatitis. *Pancreas* 2008; 36:105-12. [PMID 18376299]
3. Gupta R, Munoz JC, Garg P, Masri G, Nahman NS Jr, Lambiase LR. Mediastinal pancreatic pseudocyst--a case report and review of the literature. *MedGenMed* 2007; 9:8. [PMID 17955064]
4. Wechalekar M, Falodia S, Gamanagatti S, Makharia GK. An extension of pancreatic pseudocysts in the neck. *Pancreas* 2007; 34:171-3. [PMID 17198207]
5. Farman J, Kutcher R, Dallemand S, Lane FC, Becker JA. Unusual pelvic complications of a pancreatic pseudocyst. *Gastrointest Radiol* 1978; 3:43-5. [PMID 669165]
6. Wang SJ, Chen JJ, Changchien CS, Chiou SS, Tai DI, Lee CM, et al. Sequential invasions of pancreatic pseudocysts in pancreatic tail, hepatic left lobe, caudate lobe, and spleen. *Pancreas* 1993; 8:133-6. [PMID 8419901]
7. Mofredj A, Cadranet JF, Dautreux M, Kazerouni F, Hadj-Nacer K, Deplaix P, et al. Pancreatic pseudocyst located in the liver: a case report and literature review. *J Clin Gastroenterol* 2000; 30:81-3. [PMID 10636217]
8. Balzan S, Kianmanesh R, Farges O, Sauvanet A, O'toole D, Levy P, et al. Right intrahepatic pseudocyst following acute pancreatitis: an unusual location after acute pancreatitis. *J Hepatobiliary Pancreat Surg* 2005; 12(2):135-37. [PMID 15868077]
9. Les I, Córdoba J, Vargas V, Guarner L, Boyé R, Pineda V. Pancreatic pseudocyst located in the liver. *Rev Esp Enferm Dig* 2006; 98: 616-20. [PMID 17048998]
10. Shibasaki M, Bandai Y, Ukai T. Pancreatic pseudocyst extending into the liver via the hepatoduodenal ligament: a case report. *Hepatogastroenterology* 2002; 49:1719-21. [PMID 12397775]
11. Aiza I, Barkin JS, Casillas VJ, Molina EG. Pancreatic pseudocysts involving both hepatic lobes. *Am J Gastroenterol* 1993; 88:1450-2. [PMID 8362849]
12. Rao MS, Bendayan M, Kimbrough RD, Reddy JK. Characterization of pancreatic-type tissue in the liver of rat induced by polychlorinated biphenyls. *J Histochem Cytochem* 1986; 34(2):197-01. [PMID 2418098]

13. Tosh D, Shen CN, Slack JM. Conversion of pancreatic cells to hepatocytes. *Biochem Soc Trans* 2002; 30:51-5. [PMID 12023823]
 14. Terada T, Nakanuma Y. Expression of pancreatic enzymes (alpha-amylase, trypsinogen, and lipase) during human liver development and maturation. *Gastroenterology* 1995; 108:1236-45. [PMID 7535276]
 15. Gautier-Benoit C, Luez J, Cécile JP. Pseudocyst of the pancreas with intrahepatic development. *Sem Hop* 1974; 50:1235-7. [PMID 4372705]
 16. Hospitel S, Guinot B, Teyssou H, Meyblum J, Tessier JP. Intrahepatic localization of a pancreatic pseudocyst. *J Radiol* 1983; 64:355-8. [PMID 6876020]
 17. Lederman E, Cajot O, Canva-Delcambre V, Ernst O, Notteghem B, Paris JC. Pseudocysts of the left liver: uncommon complication of acute pancreatitis. *Gastroenterol Clin Biol* 1997; 2:340-1. [PMID 9208003]
 18. Atienza P, Couturier D, Grandjouan S, Guerre J, Bettan L, Chapuis Y, Vasile N. Intrahepatic liquid collections of pancreatic origin. One case and a review of the literature. *Presse Med* 1987; 16:1195-8.
 19. Ryu JK, Woo SM, Hwang JH, Jeong JB, Yoon YB, Park IA, et al. Cyst fluid analysis for the differential diagnosis of pancreatic cysts. *Diagn Cytopathol* 2004; 31:100-05. [PMID 15282721]
 20. Murphy BJ, Casillas J, Ros PR, Morillo G, Albores-Saavedra J, Rolfes DB. The CT appearance of cystic masses of the liver. *Radiographics* 1989; 9:307-22. [PMID 2538868]
 21. Aghdassi A, Mayerle J, Kraft M, Sielenkämper AW, Heidecke CD, Lerch MM. Diagnosis and treatment of pancreatic pseudocysts in chronic pancreatitis. *Pancreas* 2008; 36:105-12. [PMID 18376299]
 22. Scappaticci F, Markowitz SK. Intrahepatic pseudocyst complicating acute pancreatitis: imaging findings. *AJR Am J Roentgenol* 1995; 165:873-4. [PMID 7676984]
 23. Shinozuka N, Okada K, Torii T, Hirooka E, Tabuchi S, Aikawa K, et al. Endoscopic pancreatic duct drainage and stenting for acute pancreatitis and pancreatic cyst and abscess. *J Hepatobiliary Pancreat Surg* 2007; 14:569-74. [PMID 18040622]
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