CASE STUDY IN GASTROENTEROLOGY & HEPATOLOGY

Serous Microcystic Adenoma of the Pancreas

Diego Vázquez Saldaña, MD¹ David Antonio Mateo de Acosta Andino, MD² Juan Pablo Gurria, MD³ ¹Division of Colorectal Surgery, Hospital General de México, Secretaría de Salud Federal, Mexico City, Mexico; ²Division of Plastic Surgery, University of Alabama at Birmingham, Birmingham, Alabama; ³Department of Surgery, University of Illinois College of Medicine at Peoria, Peoria, Illinois

ystic pancreatic neoplasms represent approximately 15% of all pancreatic tumors.¹ Serous microcystic adenomas account for up to 25% of cystic pancreatic neoplasms and 1% of all exocrine neoplasms of the pancreas.² The etiology and pathophysiology of serous microcystic adenomas are unclear.^{3,4} Benign serous microcystic adenomas are glycogen-rich cystadenomas, whereas malignant serous microcystic adenomas are considered serous cystadenocarcinomas based upon their invasive microscopic appearance.

Serous microcystic adenomas are most common in middle-aged women and are frequently diagnosed incidentally on abdominal imaging or upon surgical exploration. Less frequent presentations include obstructive jaundice, recurrent pancreatitis, upper gastrointestinal hemorrhage, duodenal ulcers, and as part of Evans syndrome or von Hippel-Lindau syndrome.⁵⁻⁸

Case Report

A 74-year-old, type 2 diabetic woman presented to the emergency department with an exacerbation of abdominal pain that had been present for 4 years. The pain was located to the midepigastric area and was associated with early satiety and distension. The patient reported that her symptoms had recently exacerbated and had become associated with gastroesophageal reflux. On physical examination, the patient was obese and distended and had a palpable left upper quadrant mass. The mass was

Address correspondence to:

not fixed, and there was no skin discoloration associated with it. The patient's complete blood count, liver function panel, amylase level, lipase level, and tumor markers were within normal limits. A computed tomography scan of the abdomen revealed a cystic mass measuring 7.2 cm \times 6.5 cm \times 6.5 cm in the body and tail of the pancreas with a 9-mm thick wall and a honeycomb appearance (Figure 1).

Upon surgical exploration, a cystic mass was encountered in the body and tail of the pancreas with dense adhesions to the splenic hilum. A distal pancreatectomy with splenectomy was performed. Macroscopically, the tumor was cystic with clear serous fluid, measured 10 cm \times 7 cm \times 5 cm, had a gray surface, and completely replaced the



Figure 1. A computed tomography scan of the abdomen with intravenous and oral water-soluble contrast in sagittal view. There is a cystic tumor in the body and tail of the pancreas that adheres to the splenic hilum without compressing its structures.

Dr David A. Mateo de Acosta Andino, Division of Plastic Surgery, University of Alabama at Birmingham, 510 20th Street South Ste 1101, Birmingham, AL 35294; Tel: 205-934-2307; E-mail: dmacosta@uabmc.edu



Figure 2. A medial cut of a pathology specimen showing ovoid gray–whitish tissue measuring $10 \text{ cm} \times 7 \text{ cm} \times 5 \text{ cm}$ with a multicystic interior and serous fluid.

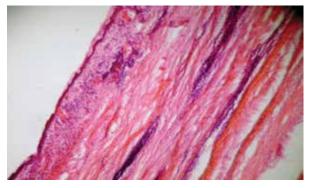


Figure 3. A microphotograph of the inner wall of the specimen (hematoxylin and eosin stain, 10× magnification). Shown is a simple cylindrical epithelium with no signs of malignancy or invasion.

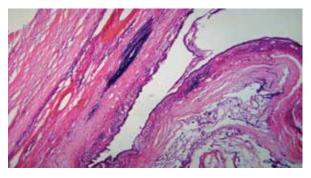


Figure 4. A microphotograph of the inner wall of the pancreatic cyst showing abundant fibroblasts and inflammatory infiltrate with vascular congestion (hematoxylin and eosin stain, 10× magnification).

body and tail of the pancreas (Figure 2). Microscopically, the inner wall of the cyst had a benign appearance with a simple cylindrical epithelium (Figure 3), areas of fibrosis, mild inflammatory reaction, and vascular congestion (Figure 4). The final diagnosis was a serous microcystic adenoma of the pancreas.

Discussion

Pancreatic serous microcystic adenomas usually have a solid appearance and an average diameter of 6 cm.⁹ These tumors may remain asymptomatic for long periods of time and are frequently found incidentally.^{10,11} The typical honeycomb appearance of these tumors is due to the multiple septums that extend from the cyst wall. Asymptomatic patients with tumors smaller than 4 cm can be followed with yearly abdominal computed tomography scans. There is still controversy on the surgical criteria for the management of this neoplasm, and some authors advocate early resection in tumors larger than 4 cm or those which are symptomatic.¹² Because of the very low malignant potential of these tumors, surgical resection, as exemplified in our patient, is only carried down to tumor-free margins.

As an alternative to surgical resection, endoscopic ultrasound ablation by ethanol injection has been used successfully in patients with small tumors to treat these lesions via a minimally invasive approach. Short-term cyst resolution has been reported in 33% to 79% of cases.¹³⁻¹⁵ DeWitt and colleagues have reported imaging-proven resolution of endoscopically ablated serous neoplasms of the pancreas for a maximum follow-up of 1 year with a complication rate of 4% to 10%.¹⁶

Summary

Serous microcystic adenomas of the pancreas are a rare entity, accounting for a very small percentage of exocrine pancreatic tumors. Nevertheless, they should be kept in the differential diagnosis. These tumors present in a nonspecific fashion, and a high index of suspicion must be kept in order to diagnose them. A computed tomography scan of the abdomen with 1-mm thick cuts following arterial, venous, and portal phase protocol typically shows a multiseptated cyst with a honeycomb appearance. Serous microcystic adenomas are surgically curable and have very limited carcinogenic risk. Due to the low malignant potential, these lesions are frequently amenable to limited local resection with distal pancreatectomy with or without splenic preservation.

The authors have no relevant conflicts of interest to disclose.

References

1. Köksal AS, Asil M, Turhan N, et al. Serous microcystic adenoma of the pancreas: case report and review of the literature. *Turk J Gastroenterol.* 2004;15(3):183-186.

2. Alsaad K, Chetty K. Serous cystic neoplasms of the pancreas. *Curr Diagn Pathol.* 2005;11:102-109.

3. Yamamoto T, Takahashi N, Yamaguchi T, Imamura Y. A case of solid variant type of pancreatic serous cystadenoma mimicking islet cell tumor. *Clin Imaging*. 2004;28(1):49-51.

4. Alasio TM, Vine A, Sanchez MA, Dardik H. Pancreatic endocrine tumor coexistent with serous microcystic adenoma: report of a case and review of the literature. *Ann Diagn Pathol.* 2005;9(4):234-238.

5. Ji Y, Wang XN, Lou WH, Sujie A, Tan YS, Jin DY. Serous cystic neoplasms of the pancreas: a clinicopathologic and immunohistochemical analysis. *Chin J Dig Dis.* 2006;7(1):39-44.

6. King JC, Ng TT, White SC, Cortina G, Reber HA, Hines OJ. Pancreatic serous cystadenocarcinoma: a case report and review of the literature. *J Gastrointest Surg.* 2009;13(10):1864-1868.

7. Galanis C, Zamani A, Cameron JL, et al. Resected serous cystic neoplasms of the pancreas: a review of 158 patients with recommendations for treatment. *J Gastrointest Surg.* 2007;11(7):820-826.

8. Hruban RH, Fukushima N. Cystic lesions of the pancreas. *Diagn Histopathol* (Oxf). 2008;14(6):260-265.

9. Targarona J, Garatea R, Romero R, et al. Tratamiento quirúrgico de los cistoadenoma serosos gigantes del páncreas reporte de dos casos. *Rev Gastroenterol Peru*. 2007;27:77-82.

10. Leonard D, Baulieux J, Rode A, et al. Multiple synchronous serous cystadenomas of the pancreas: uncommon CT and MRI findings. *J Hepatobiliary Pancreat Surg.* 2007;14(6):600-603.

11. Agarwal N, Kumar S, Dass J, Arora VK, Rathi V. Diffuse pancreatic serous cystadenoma associated with neuroendocrine carcinoma: a case report and review of literature. *JOP*. 2009;10(1):55-58.

12. Tseng JF. Management of serous cystadenoma of the pancreas. J Gastrointest Surg. 2008;12(3):408-410.

13. Oh HC, Seo DW, Lee TY, et al. New treatment for cystic tumors of the pancreas: EUS-guided ethanol lavage with paclitaxel injection. *Gastrointest Endosc*. 2008;67(4):636-642.

14. Oh HC, Seo DW, Kim SC, et al. Septated cystic tumors of the pancreas: is it possible to treat them by endoscopic ultrasonography-guided intervention? *Scand J Gastroenterol*. 2009;44(2):242-247.

15. DeWitt J, McGreevy K, Schmidt CM, Brugge WR. EUS-guided ethanol versus saline solution lavage for pancreatic cysts: a randomized, double-blind study. *Gastrointest Endosc.* 2009;70(4):710-723.

16. DeWitt J, DiMaio CJ, Brugge WR. Long-term follow-up of pancreatic cysts that resolve radiologically after EUS-guided ethanol ablation. *Gastrointest Endosc*. 2010;72(4):862-866.

(Continued from page 424)

paritaprevir, and ritonavir in an open-label study of patients with genotype 1b chronic hepatitis C virus infection with and without cirrhosis. *Gastroenterology*. 2015;149(4):971-980.e1.

 Charlton M, Everson GT, Flamm SL, et al; SOLAR-1 Investigators. Ledipasvir and sofosbuvir plus ribavirin for treatment of HCV infection in patients with advanced liver disease. *Gastroenterology*. 2015;149(3):649-659.

7. Yang JD, Aqel BA, Pungpapong S, Gores GJ, Roberts LR, Leise MD. Direct acting antiviral therapy and tumor recurrence after liver transplantation for hepatitis C-associated hepatocellular carcinoma. *J Hepatol.* 2016;65(4):859-860.

8. Foster GR, Irving WL, Cheung MC, et al; HCV Research, UK. Impact of direct acting antiviral therapy in patients with chronic hepatitis C and decompensated cirrhosis. *J Hepatol.* 2016;64(6):1224-1231.

9. Schaefer EA, Chung RT. Anti-hepatitis C virus drugs in development. *Gastroenterology*. 2012;142(6):1340-1350.e1.

10. Kwo PY, Mantry PS, Coakley E, et al. An interferon-free antiviral regimen for HCV after liver transplantation. *N Engl J Med.* 2014;371(25):2375-2382.

11. Calleja JL, Crespo J, Rincón D, et al; Spanish Group for the Study of the Use of Direct-Acting Drugs Hepatitis C Collaborating Group. Effectiveness, safety and clinical outcomes of direct-acting antiviral therapy in HCV genotype 1 infection: results from a Spanish real-world cohort. *J Hepatol.* 2017;66(6):1138-1148.

12. Ahmed Sakr A, Hanifi JM, Valerie Lin M. Successful treatment of mixed hepatitis C genotypes in a cirrhotic patient with an all-oral, interferon-free regimen. *ACG Case Rep J.* 2017;4:e16.

13. Fernández I, Muñoz-Gómez R, Pascasio JM, et al. Efficacy and tolerability of interferon-free antiviral therapy in kidney transplant recipients with chronic hepatitis C. *J Hepatol.* 2017;66(4):718-723.

14. Nault JC, Colombo M. Hepatocellular carcinoma and direct acting antiviral treatments: controversy after the revolution. *J Hepatol.* 2016;65(4):663-665.

15. Buonfigioli F, Conti F, Andreone P, et al. Development of hepatocellular carcinoma in HCV cirrhotic patients treated with direct acting antivirals. Presented at the European Association for the Study of the Liver meeting; April 13-17, 2016; Barcelona, Spain. Abstract LBP506.

 Prenner SB, VanWagner LB, Flamm SL, Salem R, Lewandowski RJ, Kulik L. Hepatocellular carcinoma decreases the chance of successful hepatitis C virus therapy with direct-acting antivirals. *J Hepatol.* 2017;66(6):1173-1181. 17. Reig M, Mariño Z, Perelló C, et al. Unexpected high rate of early tumor recurrence in patients with HCV-related HCC undergoing interferon-free therapy. *J Hepatol.* 2016;65(4):719-726.

18. Affronti A, Ju M, Catt J, Rosenberg WM, Macdonald D. Successful hepatitis C treatment in advanced cirrhosis with DAA reduces HCC incidence. Presented at the American Association for the Study of Liver Diseases meeting; November 11-15, 2016; Boston, Massachusetts. Abstract 944.

19. Nagaoki Y, Aikata H, Kobayashi T, et al. Hepatocellular carcinoma development in hepatitis C virus patients who achieved sustained viral response by interferon therapy and direct anti-viral agents therapy. Presented at the American Association for the Study of Liver Diseases meeting; November 11-15, 2016; Boston, Massachusetts. Abstract 860.

20. Muir A, Buti M, Nahass R, et al. Long-term follow-up of patients with chronic HCV infection and compensated or decompensated cirrhosis following treatment with sofosbuvir-based regimens. Presented at the American Association for the Study of Liver Diseases meeting; November 11-15, 2016; Boston, Massachusetts. Abstract 880.

21. Flemming J, Kim WR, Brosgart CL, Terrault NA. Reduction in liver transplant wait-listing in the era of direct acting anti-viral therapy. Presented at the American Association for the Study of Liver Diseases meeting; November 11-15, 2016; Boston, Massachusetts. Abstract LB-23.

22. ANRS Collaborative Study Group on Hepatocellular Carcinoma. Lack of evidence of an effect of direct-acting antivirals on the recurrence of hepatocellular carcinoma: data from three ANRS cohorts. *J Hepatol.* 2016;65(4):734-740.

23. Chokkalingam AP, Singer AW, Osinusi AO, Brainard DM, Telep L. Risk of incident liver cancer following HCV treatment with sofosbuvir-containing regimens. Presented at the American Association for the Study of Liver Diseases meeting; November 11-15, 2016; Boston, Massachusetts. Abstract 739.

24. Zucman-Rossi J, Villanueva A, Nault JC, Llovet JM. Genetic landscape and biomarkers of hepatocellular carcinoma. *Gastroenterology*. 2015;149(5):1226-1239.e4.

25. Villani R, Facciorusso A, Bellanti F, et al. DAAs rapidly reduce inflammation but increase serum VEGF level: a rationale for tumor risk during anti-HCV treatment. *PLoS One.* 2016;11(12):e0167934.