The Korean Journal of Pathology
2004; 38: 423-6

Macrocystic Form of Serous Cystadenoma of the Pancreas
– Two Cases Report –

Ki-Seok Jang · Hyo Jin Lee[1] Moon Hyang Park

Department of Pathology, College of Medicine, Hanyang University, Seoul; [1]Department of Pathology, National Police Hospital, Seoul, Korea

Received : October 7, 2003
Accepted : September 6, 2004

Corresponding Author
Moon Hyang Park, M.D.
Department of Pathology, Hanyang University, College of Medicine, 17 Haengdang-dong, Seongdong-gu, Seoul 133-791, Korea
Tel: 02-2290-8249
Fax: 02-2296-7502
E-mail: parkmh@hanyang.ac.kr

The macrocystic form of serous cystadenoma of the pancreas is an uncommon benign neoplasm composed of few, relatively large cysts that are lined by uniform, glycogen-rich, cuboidal epithelial cells. We report here on two cases of pathologically proven macrocystic serous cystadenoma of the pancreas in a 45-year-old female patient and a 53-year-old female patient. Both these cysts were lined by low cuboidal epithelia without any evidence of mucin production. There was also no evidence of pancreatitis. These tumors were radiologically suspected as being mucinous cystic neoplasm or pseudocysts. Although the microscopic and immunohistochemical studies of the macrocystic variant are not different from the conventional serous microcystic cystadenoma, their unusual macroscopic features can lead to confusion for the clinicians and radiologists.

Key Words : Cystadenoma, Serous; Pancreatic cyst

Serous cystadenomas of the pancreas are considered to be microcystic adenomas.[1] However, macrocystic variants of serous cystadenoma of the pancreas that are dissimilar to the microcystic adenomas have been described.[2] The immunohistochemical features are the same as for the serous microcystic adenomas, and this indicates that these epithelial neoplasms are probably also derived from ductal cells.[3] Serous oligocystic adenoma, which is composed of only a few relatively large cysts lined by epithelial cells, shows evidence of ductular differentiation.[4] This tumor category includes macrocystic serous cystadenoma,[2] serous oligocystic and ill-demarcated adenoma,[3] and the cystadenomas that are observed in children.[5] However, the macrocystic form of this benign tumor exhibits distinctly different macroscopic features from microcystic adenoma, and this can lead to diagnostic difficulties. Several Korean cases with their radiologic and pathologic findings have been reported in the literatures.[6-10] We report here on two additional cases of the macrocystic form of serous cystadenoma of the pancreas.

CASE REPORTS

Case 1

A 45-year-old woman was admitted to our hospital in December 2002 because of epigastric pain and nausea that had occurred for the previous 10 days. The patient had no significant past medical history such as von Hippel-Lindau disease or pancreatitis, nor did she have any history of abdominal trauma, except for an appendectomy about 20 years ago. On the laboratory findings, the results from routine complete blood count (CBC), serum biochemical studies and urinalysis were all within normal limits; carcinoembryonic antigen (CEA) and carbohydrate antigen (CA) 19-9 concentrations in the serum were both within normal limits. Abdominal computed tomography (CT) showed a homogeneous, $2.5 \times 3 \times 3$ cm sized, unilocular cystic mass in the pancreatic body that displayed hypoattenuation relative to the adjacent pancreatic parenchyma, and this lesion was without septation or an enhanced solid portion (Fig. 1A). The specimen of cystectomy

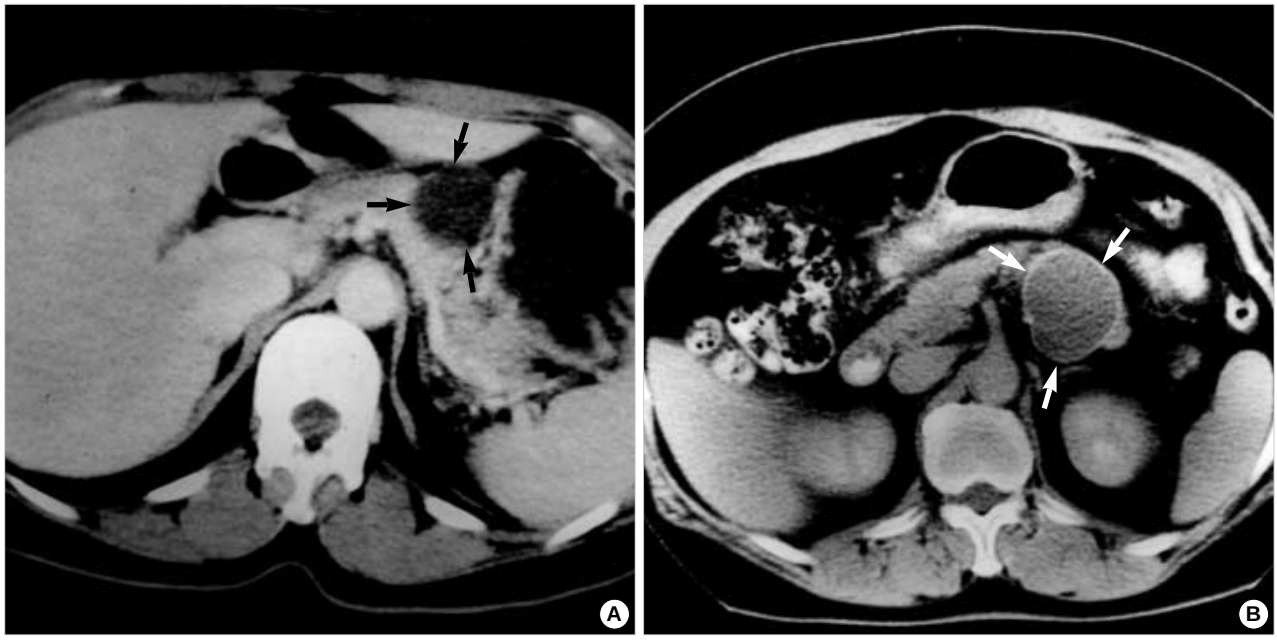

Fig. 1. (A) Abdominal computed tomography (CT) of case 1 showed a homogeneous, 2.5×3×3 cm sized, unilocular cystic mass in the pancreatic body that was hypoattenuating relative to the adjacent pancreatic parenchyma, without septation or enhanced solid portion (arrows). (B) Abdominal CT of case 2 showed a 4.5×5×5 cm sized, unilocular cystic mass in the pancreatic tail without septation or solid portion (arrows).

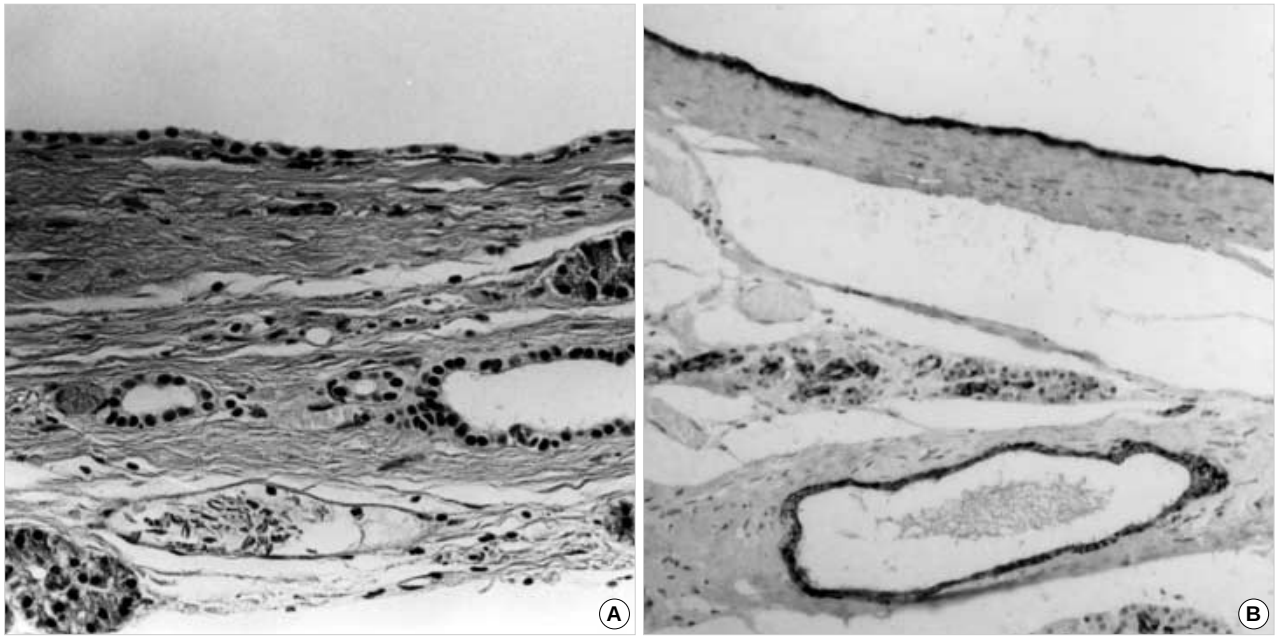

Fig. 2. Microscopic finding of case 1. (A) The cystic wall is lined by simple low cuboidal epithelium which have benign-looking central nuclei and clear or eosinophilic cytoplasm. (B) The lining epithelium is strongly immunohistochemically reactive for cytokeratin.

consisted of a pale to tan, thin-walled unilocular cyst that measured 3 cm in diameter. The inner surface was smooth without a solid portion and it contained watery fluid. Microscopically, the cystic wall was lined by simple cuboidal epithelium that was immunohistochemically positive for cytokeratin, cytokeratin 7 and cytokeratin 19, and the epithelial cells were negative for CEA. The epithelial cells had cytologically benign-appearing central nuclei and a moderate amount of clear or eosinophilic cytoplasm

without any evidence of mucin secretion. The lining epithelial cells' cytoplasm stained positively with periodic acid-schiff (PAS) and negatively with diastase-PAS, representing glycogen within the epithelial cells (Fig. 2). All these findings were consistent with the findings of the macrocystic form of serous cystadenoma. The patient is currently alive without tumor recurrence for 8 months after surgery.

Case 2

A 53-year-old woman was referred to our institution for the histologic evaluation of a 5 cm sized, unilocular cystic mass in the tail of her pancreas. The patient had no significant past medical history. The laboratory tests, including CBC and blood chemistry, were all unremarkable. Abdominal CT showed a unilocular cystic mass in the pancreatic tail that was devoid of septa or a solid portion (Fig. 1B). Microscopically, the cystic wall was lined by attenuated cuboidal epithelium that was immunohistochemically positive for cytokeratin 7, cytokeratin 19, and negative for CEA, without any evidence of mucin secretion. The cystic wall was thick and fibrotic with some compressed and rather dilated ducts, and there was a scattered infiltration of lymphocytes and plasma cells. The surrounding pancreatic tissue was well preserved.

DISCUSSION

The macrocystic form of serous cystadenoma is a uncommon benign neoplasm composed of few, relatively large cysts lined by uniform, glycogen-rich, cuboidal epithelial cells. A few cases of macrocystic serous cystadenoma have been reported, and this tumor is much less common than serous microcystic adenomas.[2,3] Most macrocystic serous cystadenomas are located in the head and body of the pancreas.[4] In the pancreas head, they may obstruct the periampullary portion of the common bile duct. The most common symptom associated with this tumor is upper abdominal discomfort or pain, and other symptoms include jaundice and steatorrhea. These neoplasia typically appear as a cystic mass with a diameter of 4-10 cm. On the cut surface, there are few (occasionally only one) macroscopically visible cysts filled with clear watery or brown fluid. Microscopically, this tumor generally has the same features as microcystic serous cystadenoma. Occasionally, however, the lining epithelium may be more cuboidal and less flattened. The cytoplasm is either clear, due to the presence of glycogen, or eosinophilic. The immunohistochemical and ultrastructural features are the same as those for serous microcystic adenoma. There is no evidence of malignant potential.[4,11]

Khadaroo, *et al*.[12] have reported a case of macrocystic serous cystadenoma that was diagnosed by a cytologic review of the cystic fluid obtained by fine needle aspiration biopsy. This diagnostic procedure can be helpful for the patient that can be managed conservatively without abdominal surgery.[12]

Macrocystic serous cystadenoma has been thought to be the variant of serous cystadenoma, and this can result in diagnostic difficulties for both the radiologists and pathologists. Several cases that were pathologically confirmed as 'macrocystic serous cystadenoma' were preoperatively diagnosed as 'mucinous cystic neoplasm' or as 'pseudocyst' by the radiologists and surgeons.[8,13,14]

The differential diagnosis includes mucinous cystic neoplasm, pseudocyst, retention cyst, congenital cyst, and enterogenous cyst. It is very important to differentiate the serous cystadenoma from the mucinous cystic neoplasm because of the malignant potential of the latter. Microscopically, the columnar epithelium of the mucinous cystic neoplasm may show cellular atypia, and it stains for mucins and CEA, while the epithelium of serous cystadenoma does not.[4] In the absence of inflammatory change and chronic pancreatitis, the pseudocyst and retention cyst can be excluded in these cases.

Therefore, the recognition of such a variant of pancreatic serous cystadenoma is very important to diagnose and properly manage the patient.

REFERENCES

1. Compagno J, Oertel JE. Microcystic Adenomas of the Pancreas (Glycogen-rich Cystadenomas): a clinicopathologic study of 34 cases. Am J Clin Pathol 1978; 69: 289-98.
2. Lewandrowski K, Warshaw A, Compton C. Macrocystic serous cystadenoma of the pancreas: a morphologic variant differing from microcystic adenoma. Hum Pathol 1992; 23: 871-5.
3. Egawa N, Maillet B, Schröder S, Mukai K, Klöppel G. Serous oligocystic and ill-demarcated adenoma of the pancreas: a variant of serous cystic adenoma. Virchows Arch 1994; 424: 13-7.
4. Solcia E, Capella C, Klöppel G. Tumors of the Pancreas, Atlas of Tumor Pathology. 3rd series , Fascicle 20. Washington, D.C.: Armed Forces Institute of Pathology, 1997; 31-9, 220-3.
5. Warfel KA, Faught PR, Hull MT. Pancreatic cystadenoma in an infant: Ultrastructural study. Pediatr Pathol 1988; 8: 559-65.
6. Cho SH, Rha SE, Byun JY, Cho SM. Mixed Microcystic and Macrocystic Serous Cystadenoma of the Pancreas: A Case Report. J Kore-

an Radiol Soc 2001; 45: 47-50.
7. Kim JH, Chung JJ, Park S, *et al*. Radiologic Findings of the Serous Cystadenomas of the Pancreas with Macrocystic or Unilocular Variants: Comparison with the Pathologic Findings. J Korean Radiol Soc 2000; 42: 107-13.
8. Jin YM, Jin H, Choi IJ. Pancreatic serous cystadenoma mimicking pseudocyst. Yonsei Med J 1997; 38: 63-5.
9. Cho YM, Gong GY, Choe GY, Yu ES, Lee IC. Serous Cystadenoma of the Pancreas. Korean J Pathol 1994; 28: 522-7.
10. Huh JR, Chi JG, Jung KC, Choe KJ, Yoon YB. Macrocystic serous cystadenoma of pancreas: a case report. J Korean Med Sci 1994; 9: 78-85.
11. Hamilton SR, Aaltonen LA. WHO Classification of Tumours, Pathology and Genetics, Tumours of the Digestive System. International agency for research on cancer, 2000; 232-3.
12. Khadaroo R, Knetman N, Joy S, Nguyen GK. Macrocystic serous adenoma of the pancreas. Pathol Res Pract 2002; 198: 485-8.
13. Fujiwara H, Ajiki T, Fukuoka K, Mitsutsuji M, Yamamoto M, Kuroda Y. Macrocystic serous cystadenoma of the pancreas. J Hepatobiliary Pancreat Surg 2000; 7: 92-6.
14. Gouhiri M, Soyer P, Barbagelatta M, Rymer R. Macrocystic serous cystadenoma of the pancreas: CT and endosonographic features. Abdom Imaging 1999; 24: 72-4.