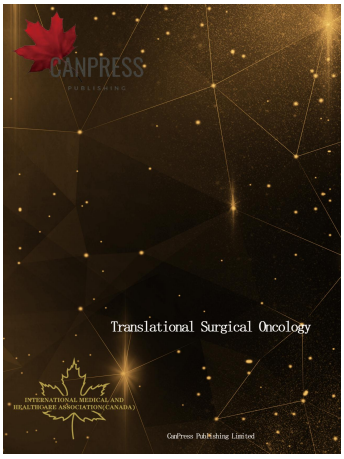
Pancreatic serous cystadenoma misdiagnosed as pancreatic neuroendocrine tumor: a case report



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Abstract

Pancreatic serous cystadenoma (SCA) appears as a multilocular mass filled with clear fluid, spongy in section, mostly solitary, and microscopically the cystic lumen is covered with flat or cuboidal epithelium. A 44-year-old woman complained of epigastric bloating for about 1 year, with worsening symptoms in the last 1 month, combined with decreased appetite and abdominal pain, and her weight had lost 3 kilograms in the last month. Computed Tomography (CT) scan showed a mass of low density lesion in the pancreatic head, with slightly unclear boundaries, and the maximum cross-sectional area was about 6.1*4.7centimetre (cm). The arterial phase of contrast-enhanced scan was obviously heterogeneous enhancement, and there were multiple small cystic non-enhancement areas. Pancreaticoduodenectomy was performed and postoperative pathological results showed pancreatic serous cystadenoma, microcystic type. We reported this case of pancreatic SCA (microcystic type) with a rare imaging finding. The high vascular signal of this imaging manifestation will raise our awareness of this tumor.

Keywords:Pancreatic serous cystadenoma; Pancreatic microcystic serous cystadenoma; Computed Tomography

Introduction

Pancreatic cystic neoplasms (PCN) are cystic neoplastic lesions originating from the epithelial of the pancreatic ducts or mesenchymal tissue and are mostly classified as benign pancreatic neoplastic lesions, with some at risk of malignancy[1]. The development of imaging technology has promoted the detection of PCN, but it still needs careful differentiation to obtain accurate diagnosis and formulate reasonable treatment. Pancreatic serous cystadenoma (SCA) is a relatively rare cystic tumor of the pancreas, which mainly includes five subtypes[2]: microcystic SCA, macrocystic (oligocytic) SCA, solid serous adenoma, von Hippel–Lindau-associated serous cyst neoplasm, and mixed serous–neuroendocrine neoplasm, of which microcystic SCA is more common. However, the lack of typical clinical manifestations in the early stages of pancreatic microcystic SCA makes it difficult to establish an accurate diagnosis, and it is often confused with some solid tumors by imaging, as reported in previous studies[3,4]. Here, we report a case of pancreatic microcystic SCA with an extremely rare imaging manifestation.

Case report

A 44-year-old woman presented to hospital on June 3, 2023 with the complaint of epigastric bloating for about 1 year, with worsening symptoms in the last 1 month, combined with decreased appetite and abdominal pain, and her weight had lost 3 kilograms in the last month. The patient had no coexisting conditions, no family history of pancreatic disease or cancer. After admission, serial examinations were carried out, and the tumor markers showed no positive results (Carbohydrate antigen (CA) 19-9 was 8.51U/ml, neuron-specific enolase (NSE) was 24.14ng/ml, CA125 was 6.69U/ml, Carcinoembryonic Antigen (CEA) was 0.57ng/ml). Computed Tomography (CT) scan showed a mass of low density lesions in the pancreatic head, with slightly unclear boundaries, and the maximum cross-sectional area was about 6.1*4.7cm. The arterial phase of contrast-enhanced scan was obviously heterogeneous enhancement, and there were multiple small cystic non-enhancement areas (Figure 1). In the pancreatic body and tail, the pancreatic duct was dilated, and

the junction of the lesion with the superior mesenteric vein, splenic vein and portal vein was not clear. A detailed physical examination was performed, which revealed no signs of jaundice and no palpable abdominal mass, except for mild epigastric tenderness. We initially diagnosed the patient as a pancreatic neuroendocrine tumor (pNET), and also possible for solid serous adenoma of the pancreas. Considering the large tumor tissue and its close relationship with venous confluence, we provided radical pancreaticoduodenectomy (PD) to the patient after communicating with the patient and her family on June 7, 2023. The surgery was difficult, but the process was smooth. The patient did not experience any discomfort and was discharged on the 9th day after the surgery. Interestingly, postoperative pathological results showed pancreatic serous cystadenoma, microcystic type. On gross examination, a grayish white slightly transparent solid mass can be seen, with a cross-sectional area of approximately 5.0*4.5cm. Immunohistochemical features include CgA(-), Syn(-), CD56(-),INSM1(-), SSTR2(-) (Figure 2). Through follow-up, the patient recovered well after surgery, with a high quality of life and no other complications.

Discussion

Pancreatic SCA is a benign tumor originating from the exocrine site of the pancreas and accounts for approximately 1-2% of all pancreatic exocrine tumors[5]. It is more common in middle-aged female patients. Some patients have no typical clinical manifestations, which can easily lead to misdiagnosis. Pancreatic SCA shows a good prognosis, and recurrence and metastasis are extremely rare[6]. Grossly, it usually appears as a multilocular mass filled with clear fluid, spongy in section, mostly solitary[7], and microscopically the cystic lumen covered with flat or cuboidal epithelium, with glycogen in the cytoplasm[8](Figure 2).

The most classical subtype of pancreatic SCA is the microcystic type, which typically presents on imaging as a solitary multilocular microcystic lesion, may be accompanied by central stellate calcifications on CT[9]. The preoperative CT images of this study were atypical, presenting a low-density mass on plain scan and markedly inhomogeneous enhancement in the arterial phase of the enhancement scan, with multiple small cystic areas of non-enhancement internally. This is an extremely rare imaging manifestation for microcystic pancreatic SCA, and in previous reports, solid, extremely microcystic pancreatic SCA can present this solid hypervascular tumor lesion[10-12], this rare manifestation on CT makes it difficult to distinguish from pNET and other solid tumors preoperatively, although some studies have shown that solid serous cystadenoma and pNET can be distinguished by early manifestations of dynamic contrast-enhanced CT[13]. Previous studies have reported cases where the two tumors were confused[14-16], which were treated surgically because of misleading preoperative imaging, and they mostly belonged to the solid serous adenoma, whereas the pathological type we report is the microcystic type. CT does not seem to be dominant in the diagnosis of this type tumor, while the combination of Magnetic Resonance Imaging (MRI) can increase the accuracy of diagnosis[17,18], as MRI is more sensitive to fluids, it is able to show the internal structure of the tumor more clearly. Endoscopic Ultrasonography (EUS) can also be an important diagnostic method and can be used to further define the tumor through puncture aspiration or biopsy[19,20]. Although most pancreatic SCA are benign tumors, it is important to formulate a reasonable treatment plan according to the tumor type and size, and most patients only need close follow-up for conservative treatment. The tumor tissue of the patient we reported was located in the pancreatic head, with large volume, combined with abdominal pain, weight loss and other obvious symptoms, and we gave PD.

Conclusion

We report a case of pancreatic SCA (microcystic type) with a rare imaging finding. The high vascular signal of this imaging manifestation will raise our awareness of this tumor, especially in the differentiation of pNET, and the adoption of more sophisticated investigations such as MRI and EUS will increase the detection rate of this tumor.

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Conflicts of Interest

The authors do not have any possible conflicts of interest.

Ethics approval and consent to participate

Written informed consent was obtained from the patient for publication of this clinical case report.

Declaration of competing interest

The authors do not have any possible conflicts of interest.

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Author Contributions

Chaoyu Pang:Writing-Original Draft, Resources, Visualization; Zhiyao Fan:Conceptualization, Visualization, Investigation; Hanxiang Zhan:Reviewing and Editing, Funding acquisition.

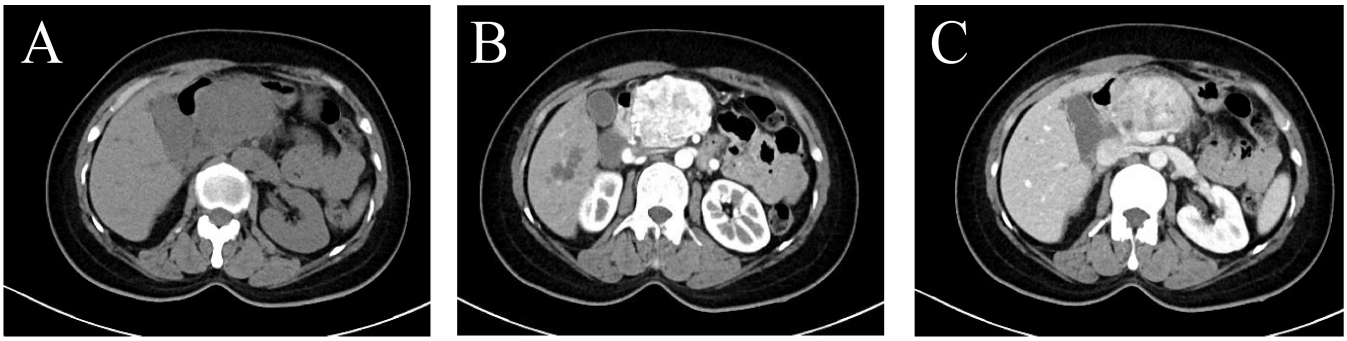

Figure 1. Contrast-enhanced CT of the abdomen.

(A) Plain CT: presenting a low-density mass. (B and C) Arterial and portal venous phase: presenting inhomogeneous enhancement with multiple small cystic areas.

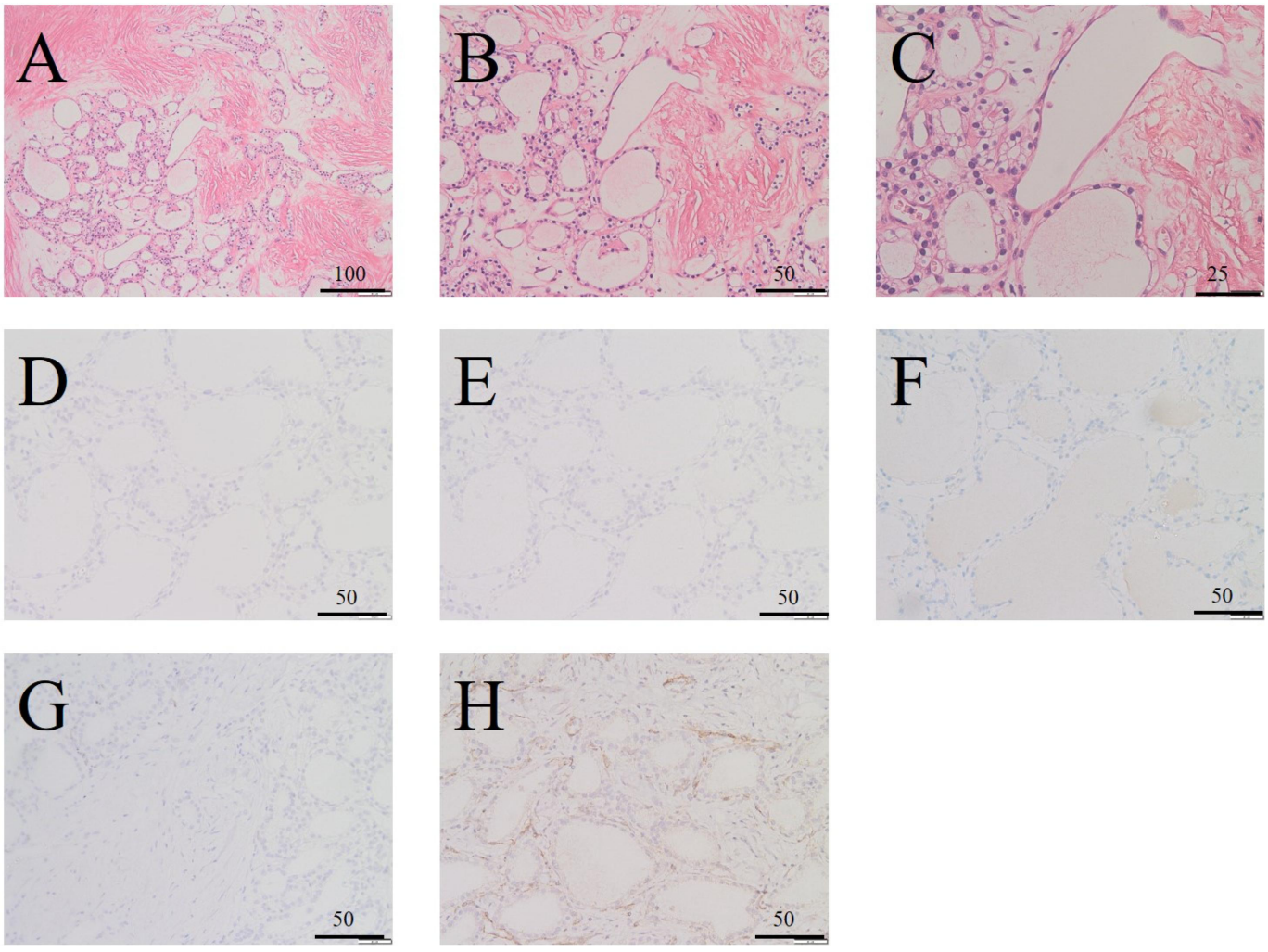

Figure 2. Pathological and immunohistochemical results.

(A-C) microscopically the cystic lumen is seen to be covered with flat or cuboidal epithelium, with glycogen in the cytoplasm, Hematoxylin-eosin staining, (D) CgA is lost in tumor, (E) Syn is lost in tumor, (F) CD56 is lost in tumor, (G) INSM1 is lost in tumor, (H) SSTR2 is lost in tumor. Original magnifications×100(A), ×200(B), and×400(C); original magnifications ×200(D-H).

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