Xanthogranulomatous Inflammation of the Peripancreatic Region Mimicking Pancreatic Cystic Neoplasm

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Abstract

Xanthogranulomatous inflammation (XGI) is histopathologically characterized by a marked proliferative fibrosis, parenchymal destruction, and infiltration of foamy histiocytes intermixed with other inflammatory cells. Herein, we report a case of a 73-year-old man without symptoms who was initially diagnosed with a pancreatic cystic tumor but later with XGI in the peripancreatic region. Although XGI has been reported to occur in various organs or tissues, such as the gallbladder, kidney, bone, stomach, colon, appendix, lymph nodes, and soft tissues, XGI involving the pancreas or its surrounding tissues is extremely rare. When a pancreatic cystic lesion does not have typical clinicoradiological features of common pancreatic cystic neoplasms, this pathologic condition should be considered in the differential diagnosis.

Key words: xanthogranulomatous inflammation, peripancreatic lesion, pancreatic cystic neoplasm


Introduction

Xanthogranulomatous inflammation (XGI) is histopathologically characterized by parenchymal destruction and marked infiltration by foamy histiocytes intermixed with other inflammatory cells (1-9). While XGI occasionally occurs in the gallbladder and kidney, XGI of the pancreas or its surrounding tissues is extremely rare (1-9). Herein, we report the case of a 73-year-old man without symptoms who was initially diagnosed with a pancreatic cystic neoplasm, but later with XGI in the peripancreatic region.

Case Report

A 73-year-old man had been diagnosed with cholecystolithiasis and acute cholecystitis at another hospital, and had undergone laparoscopic cholecystectomy. Histopathological investigation of the resected gallbladder revealed only an inflammation but not XGI. Incidentally, preoperative ultrasonography (US) and computed tomography (CT) showed a cystic lesion in the pancreatic body. Four months later, this cystic lesion remained constant in size, but the patient was referred to our hospital to undergo further investigations.

On admission, the patient was asymptomatic; he did not experience any abdominal pain, fever, weight loss, anorexia, or steatorrhea. His physical examination findings were unremarkable. He had no history of alcohol use, pancreatitis, or trauma. Biochemical laboratory data, including the level of tumor markers (alpha-fetoprotein, carcinoembryonic antigen, and CA19-9), were within the normal range. Transabdominal US revealed a 3.2×2.7-cm round, well-circumscribed mass in the pancreatic body, consisting of mainly solid and partially cystic components. Color Doppler US showed no venous flow within the mass. Contrast-enhanced CT showed that the mass was a cystic lesion encapsulated by a mild enhancing wall, which originated from the pancreatic body in abutting contact with the superior mesenteric vein (Fig. 1).

On magnetic resonance (MR) imaging, the mass appeared mildly hyperintense on T1-weighted images (Fig. 2a) and heterogeneously hyperintense, which suggested that cystic components consisting of bloody fluid or necrotic tissue debris were present in the mass (Fig. 2b). On T2-weighted MR images in the coronal plane, the mass seemed to originate from the inferior aspect of the pancreatic body (Fig. 2c). ERCP revealed a normal pancreatic duct without...
Figure 1. On contrast-enhanced CT, a cystic lesion in the pancreatic body (arrow) is revealed to be encapsulated by a thin enhancing wall.

Figure 2. MR image shows a mass with a mild hyperintensity on T1-weighted images (a) and a heterogeneous hyperintensity on T2-weighted images (b) (arrow). (c) On T2-weighted MR images in the coronal plane, the mass appears to have originated from the inferior aspect of the pancreatic body (arrow).

any communication with the mass. Fluorine-18-fluorodeoxyglucose positron emission tomography (FDG-PET) showed a ringed, pathologic uptake pattern with a maximum standardized uptake value of 3.5 (Fig. 3). On the basis of the findings of imaging studies, the differential diagnoses were considered to be solid pseudopapillary tumor and other tumors based on the cystic change which could be indicative of solid pancreatic tumors, such as islet cell tumor, acinar cell carcinoma, and ordinary ductal adenocarcinoma. Finally, the operation was commenced. A laparotomy revealed a thick-walled encapsulated mass protruding from the inferior surface of the pancreatic body, involving the serosal aspect of the third part of the duodenum and focally the portal vein. Ascites, distal metastasis, and dissemination in the abdominal cavity were not observed. Since an exact histopathological diagnosis could not be obtained through intraoperative frozen section examination, a pylorus-preserving total pancreatectomy with segmental resection of the portal vein was performed in order to achieve a radical resection.

Gross inspection of the resected specimen revealed that the mass was round and firm, measuring approximately 40 mm in diameter. The mass was found to originate not from the pancreas but from the contiguous soft tissue. The cut surface showed a cystic yellow-tan mass surrounded by ill-defined fibrous tissue. The cystic cavity contained old hemorrhagic and necrotic tissue. On histopathological examination, marked infiltration of foamy histiocytes with clear lipid-containing cytoplasm, together with abundant lymphocytes and plasma cells, was seen on the wall of the cystic mass (Fig. 4). Numerous foamy histiocytes phagocytosed bile pigments. No neoplastic cells were found. A definitive diagnosis of XGI originating from the peripancreatic region was established.

Discussion

XGI is histopathologically characterized by a marked proliferative fibrosis, parenchymal destruction, and infiltration of foamy histiocytes intermixed with other inflammatory cells (1-9). Although XGI has been reported to occur in various organs or tissues, such as in the gallbladder, kidney, bone, stomach, colon, appendix, lymph nodes, and soft tissues (1-5), XGI involving the pancreas or its surrounding tissues is an extremely rare condition (6-9).

As observed in the case presented here, pathologic condi-
tions in the vicinity of the pancreas occasionally mimic primary pancreatic neoplasm on abdominal imaging studies such as US, CT and MR imaging, which can lead to erroneous diagnoses (10). Case reports have been published that described patients who were operated on because of a presumed diagnosis of pancreatic neoplasm that was subsequently revised to peripancreatic nodal enlargement, pseudoaneurysm, or a tumor in stomach (11-15). It is important to recognize this pitfall in order to avoid unnecessary surgery or inappropriate follow-up. To the best of our knowledge, the case report presented herein is the first one on XGI in the peripancreatic region, which presented as a neoplasm of pancreatic origin.

The precise pathogenesis of XGI is not well understood. Several possible hypotheses have been proposed including defective lipid transport, immunological disorders, reaction to a specific infectious agent of low virulence, and lymphatic obstruction (5). Xanthogranulomatous cholecystitis appears to result from the accumulation of histiocytes for the phagocytosis of the extravasated bile in the gallbladder wall, which occurs secondary to the rupture of Rokitansky-Aschoff sinuses and mucosal ulceration (1, 2). There have been reports on XGI of the tissues surrounding the biliary tract in the liver and periampullary region, caused by bile leakage following gallbladder surgery and papillotomy (6). Moreover, in the case described here, it is speculated that XGI may have resulted from an inflammatory response to extravasated bile around the pancreas, because histopathological examination revealed that foamy histiocytes phagocytosed bile pigments. XGI can be induced by the spillage of bile during laparoscopic cholecystectomy (16), but the lesion in our case was already identified before the operation. It is not clear how bile had leaked into the tissues surrounding the pancreas in our case.

Since preoperative US and MR imaging in the present case revealed the cystic lesion with solid components arising from the pancreas, solid pseudopapillary tumor and the cystic change of solid pancreatic tumors, such as islet cell tumor, acinar cell carcinoma, and ordinary ductal adenocarcinoma, were considered as a differential diagnosis. Solid pseudopapillary tumor is histopathologically characterized by alternating solid and cystic components owing to degenerative changes of the solid portion (17). MR imaging characteristics of solid pseudopapillary tumor have been described to be heterogeneous high or low signal intensity on T1-weighted images and heterogeneous high intensity on T2-weighted images (17). The case we described had similar MR imaging characteristics, however solid pseudopapillary tumor develops predominantly in young women, aged 22 years on average (18). Islet cell tumor typically shows a solid pattern, but the cystic formation within the tumor occurs in 5-10% of islet cell tumors (32). Usually, it is unicellular and occupies the majority of the tumor (19-21). On contrast-enhanced CT and MR imaging, cystic islet cell tumors exhibit marked enhancement in solid components of the tumor periphery, because it is a hypervascular neoplasm (21). Also, acinar cell carcinoma and ordinary ductal adenocarcinoma rarely undergo cystic change (19, 20). Acinar cell carcinoma tends to present as a voluminous tumor at presentation. Patients with acinar cell carcinoma may present with subcutaneous fat necrosis, polyarthritis, or eosinophilia, as a result of increased lipase secreted by the neoplasm (22). Ordinary ductal adenocarcinoma generally shows abnormality in the serum levels of tumor markers such as CA 19-9 or a stricture of the pancreatic duct with dilatation of the upstream duct on pancreatogram. Therefore, we were unable to confirm the diagnosis on the basis of the clinicoradiological findings. Because the above-mentioned tumors are either malignant or have malignant potential, we decided to operate. During laparotomy, it was observed that the inflammation had spread extensively. Since these findings are suggestive of an advanced carcinoma of the pancreas, total pancreatectomy with segmental resection of the portal vein was
performed. The reason such extended surgical treatment was performed is that radical surgical resection of the mass is considered to be the best treatment for patients with clinically localized malignant pancreatic tumor, offering the only chance for cure (23).

In recent years, endoscopic ultrasound-guided fine needle aspiration (EUS-FNA) has become available for the diagnosis of pancreatic cystic lesions. Some authors advocate that analysis (cytologic examination and determination of the level of carcinoembryonic antigen and amylase) of cyst fluid obtained by EUS-FNA is useful for the preoperative diagnostic assessment of pancreatic cystic lesions, hence avoiding excessive and unnecessary surgical intervention (24, 25).

However, theoretically, cystic pancreatic lesions may carry a greater risk of tumor seeding caused by EUS-FNA, in comparison with solid lesions (26). Researchers in Japan emphasize that EUS-FNA should not be performed for patients who are suspected to have malignant cystic lesions of the pancreas on the basis of the radiological findings, as in the present case.

This report describes a case of XGI of the peripancreatic region masquerading as a pancreatic neoplasm. When a pancreatic cystic lesion does not have typical clinicoradiological features of common pancreatic cystic neoplasms, this pathologic condition should be taken into consideration in the differential diagnosis.

References


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