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Case Report

Xanthogranulomatous pancreatitis mimicking potentially malignant pancreatic neoplasm: report of a case

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Xanthogranulomatous pancreatitis (XGP) is a rare benign disease that may mimic or accompany other pancreatic diseases. Here we report a case of XGP initially suspected as malignant cystic neoplasm of the pancreas. A 64-year-old man had been incidentally found to have hypodense lesion at the body of pancreas during a lung cancer workup. All laboratory tests were within normal limits except that carcinoembryonic antigen was elevated to 31.3 ng/ml. Imaging study showed 1.8 cm sized, well demarcated, and low-attenuated mass on computed tomography (CT) with heterogeneously high intensity on T2-weighted images of magnetic resonance imaging (MRI). Under the impression of pancreas cystic neoplasm as a rare case of male solid-pseudopapillary tumor or pancreatic metastasis of lung cancer, laparoscopic distal pancreatectomy was performed. Microscopically, the mass had many foamy histiocytes with cholesterol clefts, consistent with xanthogranulomatous inflammation. Therefore, it is important to consider XGP in the differential diagnosis of pancreatic diseases. **(Ann Hepatobiliary Pancreat Surg 2017;21:243-246)**

Key Words: Xanthogranulomatous; Inflammation; Pancreatitis; Neoplasms

INTRODUCTION

Xanthogranulomatous inflammation is a rare idiopathic condition characterized by the aggregation of foamy histiocytes at various locations of the body along with chronic inflammation. These changes are found relatively common in the gallbladder and kidney. However, they are extremely rare in the pancreas.[1-3] Including the present case, only 16 cases of xanthogranulomatous pancreatitis (XGP) have been reported previously in literatures written in English. Here we report a case of XGP initially suspected as malignant cystic neoplasm of the pancreas.

CASE

A 64-year-old man was admitted to the Surgical Department for further examination of a 1.8-cm sized hypodense lesion incidentally found at the body of pancreas. He was under examination for a 3.6-cm sized lung mass at the left upper lobe which was diagnosed as moderately differentiated adenocarcinoma after left upper lobe lobectomy. The patient was a 44 pack-year smoker with diabetes requiring hyperglycemic agents. He had no reported history of alcohol intake, gallstones, or abdominal trauma.

His carcinoembryonic antigen (CEA) was elevated to 31.3 ng/ml. Liver function tests were normal. His serum levels were: AST, 29 IU/L; ALT, 24 IU/L; alkaline phosphatase, 71 IU/L; total bilirubin, 0.81 mg/dl;. amylase, 49 IU/L; C-reactive protein (CRP), 2.78 mg/dl; and hemoglobin A1c level, 6.9%. CT scan showed a 1.8-cm sized and low-attenuated mass at the body of pancreas with upstream pancreatic duct dilatation (Fig. 1A). MRI demonstrated heterogeneously high intensity on T2-weighted images and low intensity on T1-weighted images (Figs. 1B and C). The mass was well demarcated from the surrounding pancreas tissue. There was no apparent peripancreatic fat tissue involvement or lymph node enlargement. Positron emission tomography-computed tomography (PET-CT) scan showed hypermetabolic mass at the upper lobe of the

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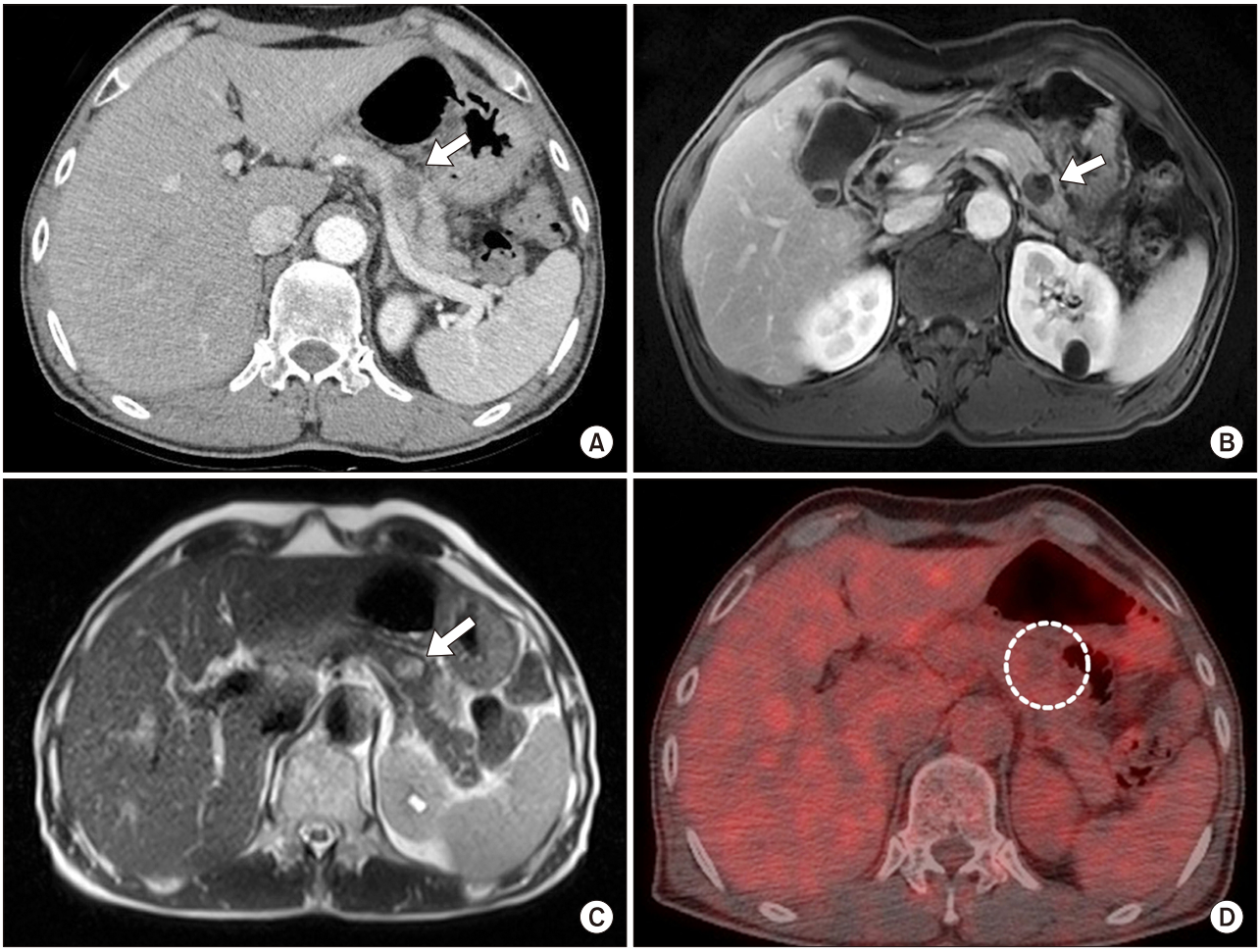

Fig. 1. (A) Axial section of CT scan showing a round 1.8-cm sized cystic mass with upstream pancreatic duct dilation (arrow). (B) MRI showing a lesion with heterogeneous low intensity on T1-weighted images (arrow). (C) MRI showing a heterogeneous high intensity on T2-weighted images (arrow). (D) PET-CT showing no significant FDG uptake.

left lung, the known lung cancer lesion site. However, no significant 2-[^{18}F]-fluoro-2-deoxy-d-glucose (FDG) uptake was found at the pancreas lesion (Fig. 1D). Endoscopic ultrasound-guided fine needle aspiration was performed for pathological diagnosis. However, it failed due to inadequately retrieved specimen for determination. Under the impression of pancreas cystic neoplasm as a rare case of male solid-pseudopapillary tumor or pancreatic metastasis of lung cancer, laparoscopic distal pancreatectomy with splenectomy was performen on June 13, 2016.

Gross examination of resected specimen revealed a 1.6 cm×1.5 cm sized yellowish mass in the pancreatic body (Fig. 2A). Microscopically, the mass had many foamy histiocytes with cholesterol clefts (Figs. 2B and C), consistent with xanthogranulomatous inflammation. Thus, the cystic lesion was diagnosed as XGP. The patient recovered fully without any complication at two weeks after surgery. There were no signs of recurrence during seven months of follow up.

DISCUSSION

Including the present case, a total of 16 cases of XGP have been reported world-wide in literatures written in English. Among them, males were predominant (12 males and 4 females). The mean age of these patients was 59.8 year (range, 30-82). These lesions were evenly distributed throughout the pancreas in the head (4/16), body (7/16), and tail (5/16). A total of 11 cases underwent surgery with clinically suspected malignant pancreas lesion.[1-9] Among them, three cases[2,5,7] were postoperatively confirmed to be combined intraductal papillary mucinous neoplasm. One

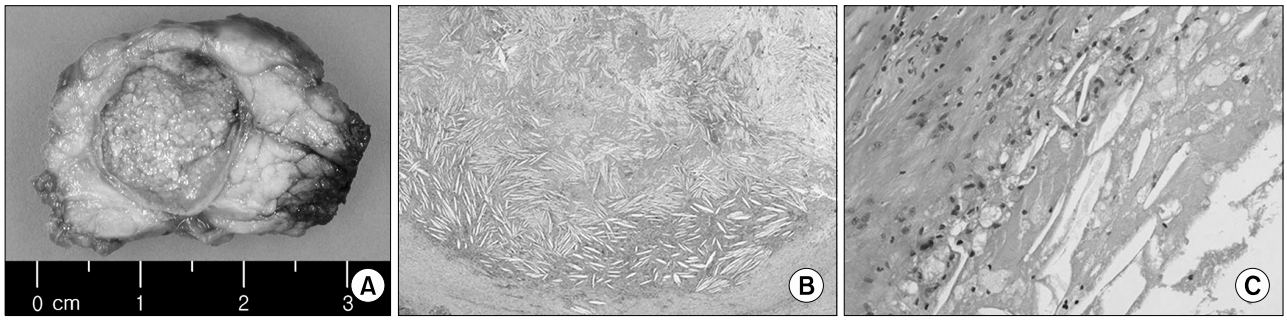

Fig. 2. (A) Gross examination of the resected specimen showing a 1.6 cm×1.5 cm sized yellowish mass. (B, C) Microscopic examination of the specimen by H&E staining showing many cholesterol clefts with foamy histiocytes (B: ×20, C: ×200).

Table 1. FDG uptake pattern of xanthogranulomatous pancreatitis lesion

Case [Ref]	Year	Sex/Age	Gross appearance	Tumor size (cm)	FDG uptake (SUV)	Uptake pattern
Ikeura, et al.[6]	2009	M/73	Cystic	4	Hot (3.5)	Ringed
Kim YN, et al.[7]	2010	F/72	Cystic	1.5×.1	Low uptake	-
Nishimura, et al.[3]	2011	M/76	Cystic	5.2×2.6	Hot (9.1)	Ringed
Kim HS, et al.[8]	2011	F/70	Solid	2.2×2.0	Hot (N/A)	N/A
Present	2016	M/60	Cystic	1.6×1.5	Low uptake	-

FDG, 2-[^{18}F]-fluoro-2-deoxy-d-glucose; SUV, standardized uptake value; N/A, not available

case[9] was combined with mucinous cystic neoplasm.

Xantogranulomatous cholecystitis (XGC) is well-recognized entity characterized by aggregation of foamy histiocytes and inflammatory cells.[4] The pathogenesis of XGC is currently unclear, although many hypotheses have been proposed. The generally accepted pathogenesis of XGC involves a series of events similar to xanthogranulomatous pyelonephritis. As described by Roberts and Parsons,[10] essential components of XGC are inflammation associated with infection, obstruction of the biliary outflow from the gall bladder due to calculi, and a source of lipid such as biliary cholesterol. Gall bladder with acute or chronic cholecystitis and partial or total obstruction of bile outflow can results in rupture of Rokitansky-Aschoff sinuses or mucosal ulceration, causing extravasation of bile into the gall bladder wall. Histiocytes accumulation in attempt to phagocytose the biliary cholesterol might result in formation of xanthoma cells. Similarly, it has been postulated that XGP might develop in the pancreas following extravasation of mucin due to obstructive changes in the pancreatic duct system.[2,5,9] With extravasation of mucin to adjacent pancreas parenchyma, inflammatory reaction might have developed. As histiocytes accumulate to phagocytose destroyed tissues, consumed adipocytes may result in xanthogranulomatous changes. In our case, the patient first developed 1.2-cm sized cyst that was increased to 1.8-cm over six months. Upstream pancreatic duct dilatation was prominent next to the mass at the body of pancreas. Grossly, it appeared as a 1.6-cm sized well-demarcated mass filled with yellowish material as if cystic lesion first developed and xanthogranulomatous material filled up the cavity later on. We believe that cystic lesion with necrotized parenchyma might have first developed through unknown reason. Over a long period of time, inflammatory change of the lesion might have caused xanthogranulomatous changes. However, the pathogenesis of XGP needs to be studied further, considering the fact that XGP is extremely rare condition compared to numerous diseases. In addition, the conditions that can cause ductal obstruction with inflammation should be considered.

XGP clinically and grossly often mimics other mass forming pancreatic lesions.[9] It is extremely difficult to distinguish XGP from pancreatic neoplasm through imaging studies.[5] Thus, radiologically it is often confused with carcinoma of the pancreas. There are no definite characteristics that can differentiate XGP from other pancreatic lesions. In our case, the lesion appeared as cystic lesion with upstream pancreatic duct dilatation. The increase in size in six months led to an impression of possible malignant cystic neoplasm. MRI showed heterogeneous high in-

tensity on T2-weighted images and heterogeneous low signal intensity on T1-weighted images, leading to the impression of solid pseudopapillary tumor. Nishimura et al.[3] have suggested that ringed uptake pattern of FDG-PET may indicate a diagnostic feature of cystic-type XGP. After reviewing six cases of XGP with FDG-PET, four cases showed hot uptake (Table 1). Among these four cystic type cases, only two cases had ringed uptake pattern. The other two cases did not show significant uptake, including the present case. They were relatively small in gross size (1.5 cm and 1.6 cm, respectively) compared to those with ringed uptake (4 cm and 5.2 cm, respectively). In comparison, two solid type cases showed hot uptake, although they were relatively small in size (2.2 cm, both). Since XGP is associated with inflammatory reaction, glucose metabolism of the lesion might be increased. Therefore, false-positive FDG uptake may result in XGP. Furthermore, ringed uptake pattern may indicate cystic type XGP. However, such findings alone are insufficient for differential diagnosis of XGP from other pancreatic diseases.

Xanthogranulomatous pancreatitis is a rare benign disease that may mimic or accompany other pancreatic diseases in imaging studies. Therefore, it is important to consider it in the differential diagnosis of pancreatic diseases. However, more cases need to be reviewed to identify unique characteristics of XGP for differential diagnosis.

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