

CASE REPORT

Xanthogranulomatous Pancreatitis Mimicking Solid Pancreatic Tumor

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ABSTRACT

Context Xanthogranulomatous lesion is a rare type of chronic inflammation, in which lipid-laden histiocytes admixed with other inflammatory cells are deposited at various locations in the body. We present a case that was treated by distal pancreatectomy and splenectomy with a clinical and radiological diagnosis of solid tumour pancreas, and was found to be xanthogranulomatous pancreatitis on histopathology. **Case report** A Sixty-five-year-old lady presented with abdominal pain and abdominal mass. Computer tomography and Ultrasound scan confirmed the presence of a soft tissue density mass in the proximal part of pancreas. With the diagnosis of a solid tumour, distal pancreatectomy was performed. Grossly the mass was solid, resembling a neuroendocrine tumour and on histopathologic study, was found to be xanthogranulomatous pancreatitis. The cells were strongly positive for CD68. **Conclusion** Although extremely rare, xanthogranulomatous pancreatitis is an important radiological differential diagnosis for solid tumour of pancreas.

INTRODUCTION

Xanthogranulomatous inflammation is a type of chronic inflammatory process characterized by infiltration of lipid-laden foamy histiocytes and other inflammatory cells. These changes have been found in the gallbladder, kidney, bone, ovary, endometrium, vagina, prostate, lymph node, and in soft tissue. However, xanthogranulomatous lesions in the pancreas are extremely rare. Most cases of Xanthogranulomatous pancreatitis were preoperatively misdiagnosed as pancreatic tumor [1]. We report a case of 65 year old female who underwent distal pancreatectomy with a clinical diagnosis of pancreatic tumour and was later diagnosed as xanthogranulomatous pancreatitis.

CASE REPORT

A Sixty-five-year-old female presented with abdominal pain, on and off for six months which was mild, dragging type and not associated with food intake. Patient also complained dysphagia and belching. She had history of loss of weight and loss of appetite. She was on regular medication for hypertension and gave a history of pulmonary tuberculosis ten years back for which she completed medication for six months. She was not diabetic. No history of alcohol intake or smoking. Her family history was uneventful.

On examination, a freely mobile mass was palpated in left hypochondrium and observed mild tenderness in epigastrium.

Her laboratory test results were as follows: complete blood count-within normal limits, total bilirubin-0.6, alkaline phosphatase-249 U/L (50-130U/L), serum amylase-142.3 U/L (22-85 U/L). Fasting and 2 hour postprandial blood sugar values were normal. Oesophagogastroduodenoscopy (OGDscopy) detected antral gastritis only. Ultrasound scan (USG) revealed a mass measuring 5.6×3.9 cm, in the body of pancreas with inhomogeneous internal echoes and suggested a differential diagnosis of 1) pancreatic mass, 2) pseudo cyst. In the computed tomography (CT) scan, identified a well defined heterogeneously enhancing soft tissue density mass, measuring 6.9×5.4×4.5 cm, in the proximal part of pancreas-consistent with malignant neoplasm of pancreas (**Figure 1**). The USG or CT scans did not show any dilated pancreatic ducts.

With the diagnosis of malignant neoplasm of the pancreas, laparotomy was performed on June 21, 2016. A large firm mass was seen in the body of the pancreas with dense fibrotic adhesions around the tumour. Distal pancreatectomy with splenectomy was performed. The patient's post-operative course was uneventful.

Grossly, identified an intact mass, measuring 7.3×7.3×6 cm, in the pancreas. The cut surface of the mass was yellowish with well circumscribed borders (**Figure 2a**). There were no areas of hemorrhage or necrosis. The adjacent pancreatic parenchyma and spleen were unremarkable. There were no dilated pancreatic ducts or calculi seen. Microscopic examination from the mass showed plenty of neutrophils (**Figure 2b**), plasma cells with numerous Russell bodies, foamy histiocytes, and lymphocytes. There was no pancreatic tissue within the mass. No epitheloid granuloma, with or without caseous necrosis, were identified. The mass-parenchyma interphase showed fibrosis with acinar atrophy and little

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Figure 1. Soft tissue density mass in CT scan.

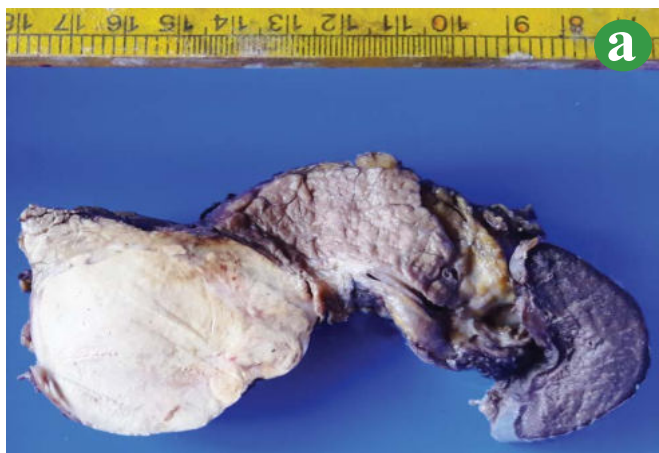


Figure 2a. Well circumscribed mass in proximal part of pancreas with yellowish white cut surface. Adjacent pancreatic parenchyma is unremarkable.

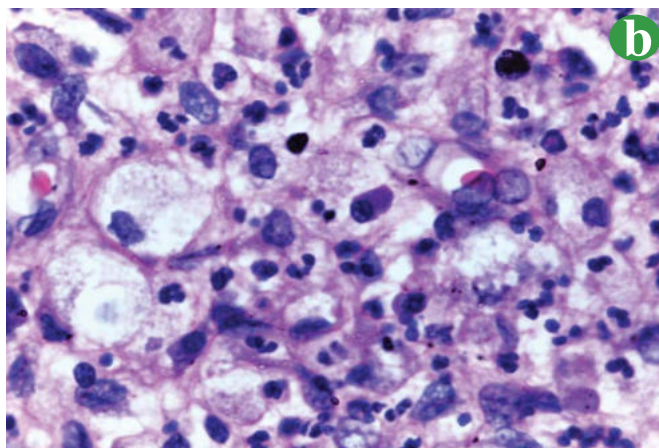


Figure 2b. (100x) -showing plenty of neutrophils, foam cells, plasma cells and lymphocytes

inflammation. Rest of the pancreatic parenchyma was unremarkable with no inflammation, necrosis or duct pathology. Immunohistochemical study of the large cells showed strong positive reaction for CD68 (Figure 2c) and negative for cytokeratin, ALK-1, CD30 and SMA, confirming its histiocytic origin.

DISCUSSION

Xanthogranulomatous inflammation is a type of chronic inflammatory reaction, microscopically characterized by foamy histiocytes and infiltration of various inflammatory

cells. This type of reaction is well established in gall bladder and kidney, and may also occur in skin, retroperitoneum, gastrointestinal tract, genital organs, lymph nodes, and soft tissue [1, 2, 3, 4, 5, 6], but rare in pancreas. Predisposing conditions proposed for xanthogranulomatous changes in the gall bladder are increased intramural pressure due to tumour or gall stones, leading to extravasation of bile and mucin and result in xanthogranulomatous changes [7]. In the kidney it may be induced by extravasation of urine [8]. It starts as acute inflammatory process which later progress to granulomatous reaction and then a cellular-type immune response [2]. Grossly, the lesion is ill circumscribed with brown or yellow nodular appearance. The lesions are more common in females and can be unilateral or bilateral [7]. Ultrastructurally, foamy macrophages initially contain bacteria and subsequently contain numerous phagolysosomes. Escherichia coli is the usual etiologic agent, but proteus mirabilis and staphylococcus aureus have also been found to be responsible [9].

Xanthogranulomatous pancreatitis (XGP) is extremely rare. Only 12-20 cases have been reported so far and most of them are from Asian countries. The peculiarity of this lesion is that, in almost all cases they were misdiagnosed as malignancy clinically and radiologically, as they presented as mass and showed local tissue invasion and destruction [2], and were treated with pancreatectomy. In contrast to xanthogranulomatous pyelonephritis, majority of XGP occurred in males [3]. The most frequent symptom was upper abdominal pain. Kim et al. [6] has done excellent review of characteristics of all cases reported so far. Three of those cases were associated with intraductal papillary mucinous neoplasm (IPMN) [1, 2, 3]. Pathogenesis of XGP is not well known, though several hypotheses have been suggested. According to Kim et al. [2] increased intraductal pressure and subsequent leak of mucin into pancreatic parenchyma by IPMN, is the cause for XGP. Kamitani et al. [1] reported that IPMN caused impairment of the passage of pancreatic juice and increased intraductal pressure, which caused leakage of mucin and induced XGP. Iyer et al. [4] suggests that XGP may be caused by obstruction of pancreatic ducts by stone followed by secondary bacterial infection. Ueno et al. [10] reported xanthogranulomatous inflammation in the wall of a pancreatic pseudocyst,

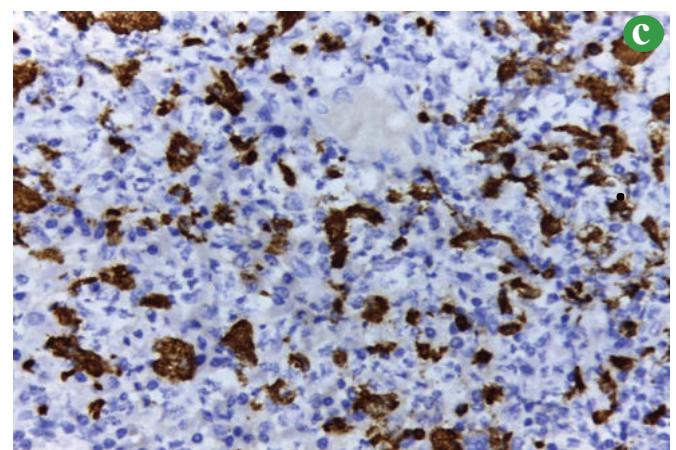


Figure 2c. (40x) - showing strong CD68 positivity.

probably due to infection and hemorrhage in the cyst. However, in our case, no such obstructive etiology was identified.

CONCLUSION

Most cases of XGP mimic primary pancreatic neoplasm. As it is difficult to differentiate XGP from true neoplastic lesions radiologically, it is important for pathologists, radiologists, and clinicians to recognize this uncommon entity so as to prevent unnecessary surgery and patient morbidity.

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Conflict of Interest

The authors have no conflicts of interest.

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