CASE REPORT

Xanthogranulomatous pancreatitis mimicking a malignant solid tumour

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SUMMARY

Xanthogranulomatous inflammation, which is known to occur in several viscera, is rarely found to affect the pancreas. We report a case of xanthogranulomatous pancreatitis (XGP) occurring in a 60-year-old man who presented with epigastric pain and vomiting. Physical examination did not reveal any abnormality. Contrast-enhanced CT of the abdomen revealed an ill-defined, heterogeneous mass lesion in the uncinate process of the pancreas, suggestive of malignancy. Whipple's pancreaticoduodenectomy was performed and the final pathological diagnosis was XGP. The patient's postoperative course was uneventful. When a pancreatic mass does not show clinico-radiological features typical of common pancreatic neoplasms, XGP should be considered for a differential diagnosis and duodenum preserving surgery can be considered.

BACKGROUND

The occurrence of xanthogranulomatous inflammation (XGI) in the pancreas is extremely rare. Only 14 cases of xanthogranulomatous pancreatitis (XGP) have been reported to date. The presented report is the third case from India.

CASE PRESENTATION

A 60-year-old man presented with epigastric pain radiating to the back for 1 month and three episodes of non-bilious vomiting in the last 1 week. He had lost 5 kg over the last 3 months. His appetite was normal. He underwent laparoscopic cholecystectomy 1 year earlier for symptomatic gallstone disease. The histopathology of the gallbladder was reported as chronic follicular cholecystitis. Physical examination showed no abnormality.

INVESTIGATIONS

CT of the abdomen showed an ill-defined lesion with predominant irregular peripheral enhancement and internal necrotic areas having possible continuity with the uncinate process of the pancreas and infiltration of the superior wall of the third and fourth parts of the duodenum (figure 1). CT guided fine needle aspiration cytology of the mass was suspicious of malignancy. Serum carcinoembryonic antigen was 2.9 ng/mL and CA 19.9 was <0.60 U/mL. Serum amylase and serum lipase were 64 and 34 U/L, respectively.

TREATMENT

The patient underwent Whipple's pancreaticoduodenectomy. Intraoperatively, growth was found in

Figure 1 Ill-defined heterogeneous mass lesion with predominant irregular peripheral enhancement and internal necrotic areas with possible continuity with the uncinate process.

the uncinate process of the pancreas, not infiltrating the SMA, superior mesenteric artery/SMV, superior mesenteric vein (SMA/SMV)/portal vein.

Gross examination of the pancreas showed a large area of necrosis surrounded by yellowish-white areas (figures 2 and 3). Microscopic examination showed areas of necrosis surrounded by sheets of foamy macrophages and chronic inflammatory cells with foreign body giant cells. Surrounding pancreatic parenchyma showed atrophy and inflammatory atypia of acinar cells (figures 4 and 5), suggesting a diagnosis of XGP.

Figure 2 Gross specimen of Whipple's resection for suspected pancreatic carcinoma: Inset—shows pancreatic parenchyma (black arrow) with a large irregular area of necrosis and yellowish-white areas of xanthogranulomatous inflammation (red arrow).

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Figure 3 Photomicrograph showing large area of necrosis above and dense inflammation destroying the pancreatic parenchyma below, H&E, $\times 100$. Inset—photomicrograph showing dense inflammation composed of neutrophils, sheets of foamy macrophages and occasional foreign body giant cells, H&E, $\times 200$.

An accessory right hepatic artery was found to arise from the SMA (video 1).

OUTCOME AND FOLLOW-UP

The patient's immediate postoperative period was uneventful. He was discharged on the 10th postoperative day after ensuring drain fluid amylase and bilirubin were normal. He was followed every 2 weeks on an OPD basis. He was asymptomatic and had a normal diet. Ultrasonogram of the abdomen after 1 month was normal.

DISCUSSION

XGI is a form of chronic mixed inflammatory process characterised by lipid-laden histiocyte aggregation and mass formation. Fairly large numbers of cases of xanthogranulomatous cholecystitis and pyelonephritis have been reported with several hypotheses including haemorrhage, immunological conditions, reaction to a specific infectious agent of low virulence (more likely for pyelonephritis), defective lipid transport and lymphatic obstruction. Bile extravasation in the gallbladder wall secondary to the

rupture of Rokitsansky-Aschoff sinuses and mucosal ulcer is one of the proposed hypothesis for xanthogranulomatous cholecystitis.¹ The pathogenesis of XGP is not well understood. Several hypotheses have been suggested for XGP based on the risk factors and clinicopathological findings. Ueno *et al*² reported XGI in the wall of a pancreatic pseudocyst, it could have been due to infection and haemorrhage in a cyst formed by cystic neoplasm, or after pancreatitis and increased intracystic pressure, leading to XGI around the pancreatic cyst wall. Kim *et al*³ advocated increased intraductal pressure and subsequent leak of mucin into the pancreatic parenchyma due to increase mucin production by intraductal papillary mucinous tumour. Iyer *et al*⁴ reported XGP as mass lesions with a history of gall stones, which might have led to obstruction of the pancreatic duct and thus inflammatory changes, as in our case.

As described in the present case and previous reports, most cases of XGP mimic primary pancreatic neoplasms. The CT findings of xanthogranulomatous pyelonephritis are calcification, increase in renal volume, hydronephrosis and hypodense areas depicting parenchymal destruction filled with either pus or debris. This disease process shares many characteristics with a true neoplasm.⁵ CT findings in xanthogranulomatous cholecystitis show marked gallbladder wall thickening and bladder wall often containing nodules, hypoattenuating at CT, that may be

Figure 4 Small focus of calcification noted within the mass.

Video 1 An accessory right hepatic artery arising from the superior mesenteric artery.

Table 1

Author	Year	Age/sex	Symptoms	History of gall stone disease	Preoperative diagnosis	Gross appearance	Site	FNAC	Operation
Ueno ²	1993	42/M	Abdominal pain	Nil	Pseudo cyst	Cystic	Body	No	Distal pancreatectomy
Iyer ⁴	2004	50/M	Jaundice	Cholelithiasis	Malignancy	Nodular	Head	Yes	Whipple's operation
Iyer	2004	36/M	Stone	Choledocholithiasis	Malignancy	Massive	Tail	Yes	Mass excision
Kamitani	2005	82/M	Epigastric pain	Nil	IPMT	Cystic	Body	No	Whipple's operation
Kang BW	2007	22/F	Abdominal pain	Nil	Chronic pancreatitis	solid	Head	No	PPPD
Okabayshi	2007	60/M	Abdominal pain	Nil	Pseudo cyst	Cystic	Tail	No	Distal pancreatectomy
Okabayshi	2007	69/M	Abdominal pain	Nil	Pseudo cyst	Cystic	Tail	No	Distal pancreatectomy
Shima	2008	66/M	Epigastric pain	Nil	Malignancy	Solid	Body	No	Distal pancreatectomy
Iso	2008	82/M	Weight loss	Nil	IPMC	Nodular	Tail	No	Distal pancreatectomy
Ikeura	2009	73/M	Abdominal pain	Nil	SPPT	Cystic	Body	No	PPPD
Uguz	2010	30/M	Abdominal pain	Nil	Chronic pancreatitis	—	Head	No	DPP
Uguz	2010	34/F	Abdominal pain	Nil	Chronic pancreatitis	—	Head	No	DPP
Kim YN ³	2010	72/F	Abdominal pain	Nil	IPMT	Cystic	Body	No	PPPD
Kim SH ⁸	2011	70/F	Abdominal pain	Nil	Malignancy	Solid	Head	No	Whipple's operation
Present case	2015	60/M	Abdominal pain	Cholelithiasis	Malignancy	Solid	Head	Yes	Whipple's operation

DPPD, duodenum preserving pancreatectomy; FNAC, fine needle aspiration cytology; IPMC, intraductal papillary mucinous carcinoma; IPMT, intraductal papillary mucinous tumour; PPPD, pylorus preserving pancreato-duodenectomy; SPPT, solid pseudo papillary tumour.

abscesses or foci of XGI. These features overlap with those of gallbladder carcinoma, often making preoperative distinction between these entities impossible.⁶ The efficacy of fluorodeoxyglucose positron emission tomography for differentiating malignant from benign pancreatic lesions remains debatable.⁷ However, we did not perform other imaging, such as MRI and PET scan, as the patient could not afford it.

All the cases reported to date have been from Asian countries, and have had a male predominance (M:F—11:4). The location of the lesions were the head (6/15), body (5/15) and tail (4/15).

Learning points

- Xanthogranulomatous pancreatitis should be considered in the differential diagnosis of a pancreatic mass.
- Inflammatory atypia of acinar cells of the pancreas may be mistaken for malignant cells in fine needle aspiration cytology (FNAC), particularly when clinically suspicious for malignancy. Hence, final diagnosis can only be made on the histopathological sample.
- It is difficult to differentiate XP from malignancy though calcification on CT (figure 6), and a well-defined margin can be considered in favour of XP.
- Predominance in Asian males and association with gall stones can be areas of further study.
- Duodenum-preserving pancreatic head resection, which has less morbidity and mortality, can be performed if XGP is diagnosed preoperatively using fine needle aspiration cytology, or by frozen section.

Importantly, the preoperative diagnosis in 9 cases (9/15) was malignancy. Pseudo cyst and chronic pancreatitis were diagnosed in three cases each (table 1). All the cases, including ours, were treated with surgery. It is difficult to differentiate XGP from true neoplastic lesions preoperatively using imaging techniques and image-guided biopsy, as in our case. Although this type of pancreatitis is extremely rare, it is important for surgeons, pathologists and radiologists to recognise this uncommon entity.

Competing interests None declared.

Patient consent Obtained.

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REFERENCES

- Houston JP, Collins MC, Cameron I, *et al.* Xanthogranulomatous cholecystitis. *Br J Surg* 1994;81:1030–2.
- Ueno T, Hamanaka Y, Nishihara K, *et al.* Xanthogranulomatous change appearing in the pancreas cyst wall. *Pancreas* 1993;8:649–51.
- Kim YN, Park SY, Kim YK, *et al.* Xanthogranulomatous pancreatitis combined with intraductal papillary mucinous carcinoma in situ. *J Korean Med Sci* 2010;25:1814–17.
- Iyer VK, Aggarwal S, Mathur M. Xanthogranulomatous pancreatitis: mass lesion of the pancreas simulating pancreatic carcinoma—a report of two cases. *Indian J Pathol Microbiol* 2004;47:36–8.
- Kim JC. US and CT findings of xanthogranulomatous pyelonephritis. *Clin Imaging* 2001;25:118–21.
- Levy AD, Murakat LA, Abbott RM, *et al.* From the archives of the AFIP. Benign tumors and tumorlike lesions of the gallbladder and extrahepatic bile ducts: radiologic-pathologic correlation. Armed Forces Institute of Pathology. *Radiographics* 2002;22:387–413.
- Mansour JC, Schwartz L, Pandit-Taskar N, *et al.* The utility of F-18 fluorodeoxyglucose whole body PET imaging for determining malignancy in cystic lesions of the pancreas. *J Gastrointest Surg* 2006;10:1354–60.
- Kim HS, Joo M, Chang SH, *et al.* Xanthogranulomatous pancreatitis presents as a solid tumor mass: a case report. *J Korean Med Sci* 2011;26:583–6.

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