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Xanthogranulomatous pancreatitis in a male patient with pre-existing SPEN

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ABSTRACT

Background: Xanthogranulomatous (yellow granulomatous) pancreatitis is a very rare and chronically destructive inflammatory process. Solid pseudo papillary pancreatic tumors are rare pancreatic tumors. *Objective:* At the Department of Abdominal Radiology, King Faisal Specialist Hospital and Research Center in Riyadh, Saudi Arabia, we believe it is the first case of Xanthogranulomatous pancreatitis associated with an existing solid pseudo papillary epithelial tumor (SPEN). *Case report*: A 30-year-old man, medically free, with a history of gastric sleeve resection, presenting with fever, recurrent severe epigastric pain, nausea and vomiting after distal pancreatic resection and spleen resection, pancreas. Histopathological analysis of histology showed solid pseudo granulomatous pancreatitis, as well as severe yellow granulomatous pancreatitis. *Conclusions:* Our case is unique in that Xanthogranulomatous pancreatitis was found in an existing male patient with SPEN.

Keywords: Xanthogranulomatous, pancreatitis, pseudo papillary neoplasms, pancreatic tumors

1. INTRODUCTION

Xanthogranulomatous pancreatitis (XGP) is exceedingly rare condition (Kim et al., 2011). In most cases, XGP without exploration usually incorrectly diagnosed as pancreatic tumor (Becker-Weidman et al., 2017). Xanthogranulomatous pancreatitis (XGP) is the very rare reasons of a cystic pancreatic mass. The pathophysiology of this procedure is not completely clear but it may probably due to a recipe of duct occlusion, infection, and recurrent hemorrhage. However, xanthogranulomatous changes in the pancreas are extremely rare (Kwon et al., 2018).

Solid and papillary epithelial neoplasm (SPEN), are rare pancreatic tumors that account for 1-2% of exocrine pancreatic disorders. It is a low-grade, low-potential neoplasm that is common in young and adolescent women and seldom disturbs males (Minz et al., 2001). Precise diagnosis of SPEN is a principal, as SPEN is unlikely to be malignant and rapid diagnosis and surgery can increase the life expectancy of patients, including the possibility of cure. The cause of this tumor is still unknown. Due to the increasing prevalence in young women, these neoplasms are thought to originate from

either pluripotent pancreatic stem cells or cells of the female genital epithelium (Naar et al., 2017).

We report what we believe to be the first case of xanthogranulomatous pancreatitis associated with pre-existing SPEN in the abdominal radiology department, Najran University, KSA.

2. CASE REPORT

30 years old male, medically free, with past surgical history of sleeve gastrectomy, presented with fever, recurrent severe epigastric pain, nausea and vomiting. The patient was worked up on his local hospital and found to have pancreatic mass on MRI with prior non-conclusive biopsies. On physical examination the patient has soft, lax abdomen with mild tenderness over the epigastric area. WBC 3.65 x 10^9/L, CRP 80.8 mg/L, Alkaline phosphatase 66 U/L, GGT 62 IU/L, lipase 57 IU/L, amylase 28 U/L, ALT 118 U/L, AST 156.8 U/L, AFP 1.09 ug/L, CA 19-9 25.60 U/L, CA 12-5 12.2 U/L, CEA 0.4 ug/L. The outside abdominal MRI showed focal pancreatic body solid lesion showing intermediate signal intensity on T2-weighted images, diffusion restriction on DWI, as well as hypoenhancement on dynamic post-contrast images. There was upstream mild main pancreatic ductal dilatation. In addition to peripancreatic stranding of fat planes.

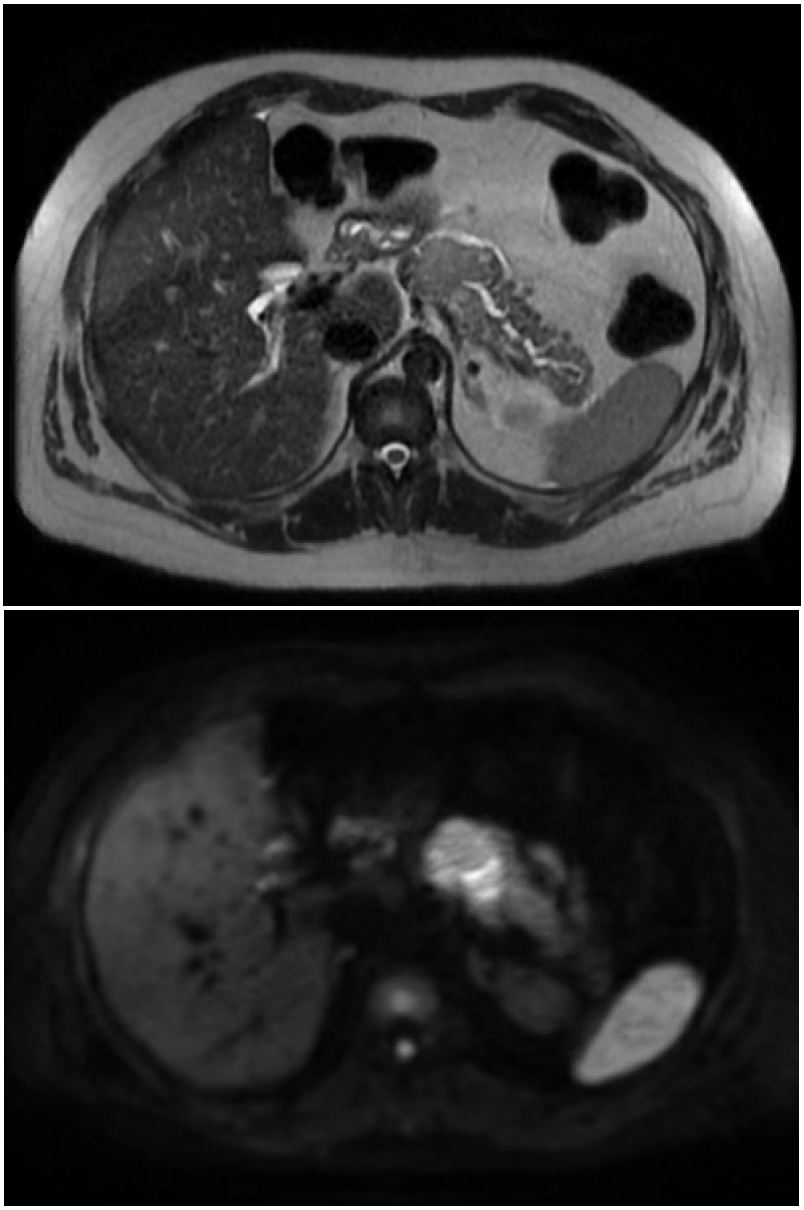

Figure 1 Outside MR study (A) axial T2 and (B) high b-value DWI; showed focal mass in the pancreatic body with intermediate signal intensity on T2-weighted images and diffusion restriction, with mild upstream main pancreatic ductal dilatation. Note the minimal stranding of the adjacent fat planes.

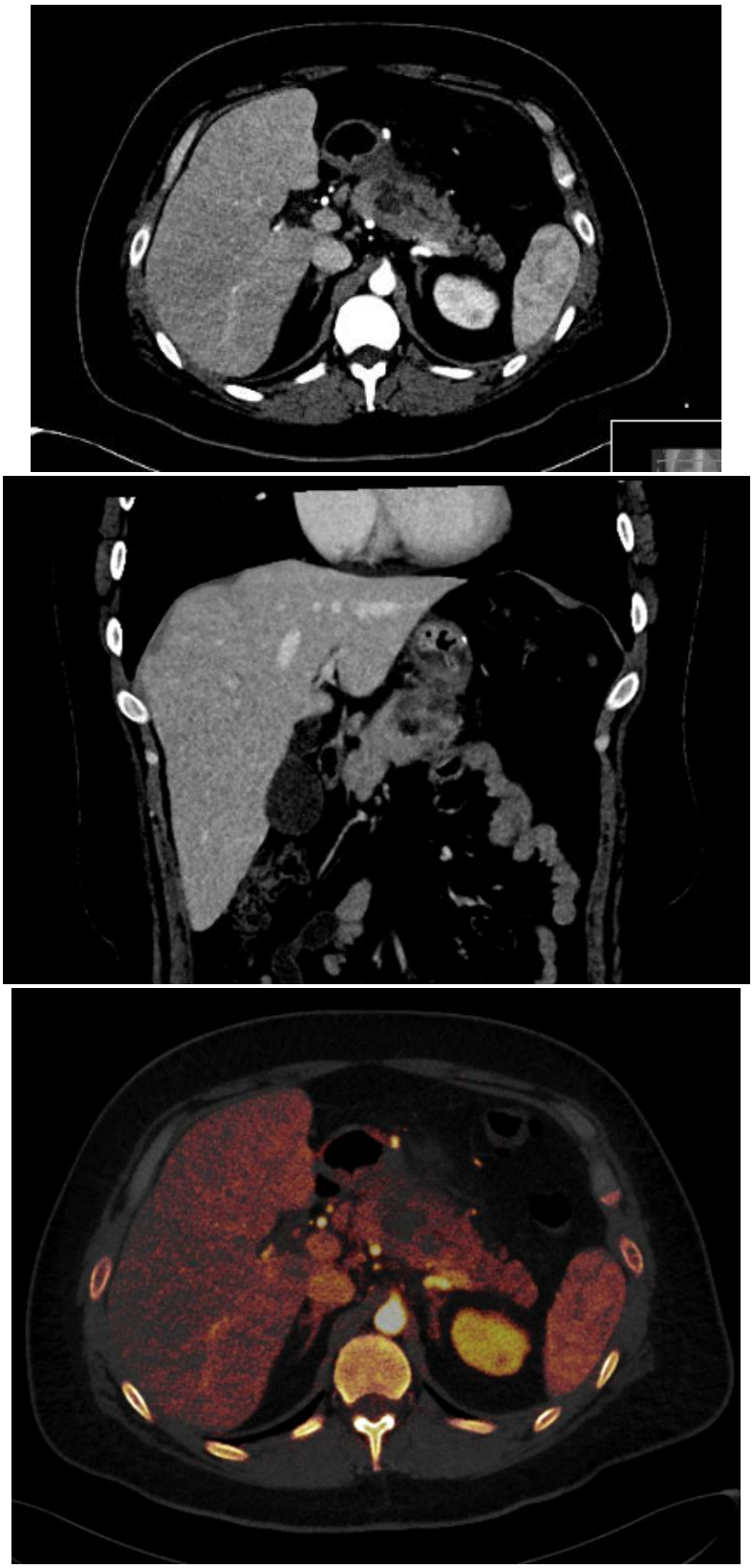

Figure 2 Dual energy contrast enhanced CT study (A) axial late arterial image, (B) coronal late arterial image, (C) axial iodine map image of the upper abdomen showing the interval morphologic change of the mass being currently of predominantely cystic component. Notice the hypoenhancement of the lesion and extension into the adjacent gastric wall.

Then the patient was suspected to have pancreatic tumor and CT, MRI and PET/CT were performed at our institution. Imaging showed interval morphologic changes of the pancreatic solid lesion, being currently of predominant cystic component, with worsening of the upstream main pancreatic ductal dilatation. The lesion is now seen extending to the adjacent greater gastric

curvature with minimal gastric wall thickening and enhancement. There is interval worsening of stranding of the adjacent fat planes. No upper abdominal lymphadenopathy. PET/CT showed avid FDG uptake in the pancreatic lesion.

The patient was diagnosed to have necrotic pancreatic mass for which he underwent distal pancreatectomy and splenectomy. Histopathological analysis of the pancreatic tissue showed severe Xanthogranulomatous pancreatitis in addition to solid pseudopapillary neoplasm (figure 1, 2 & 3).

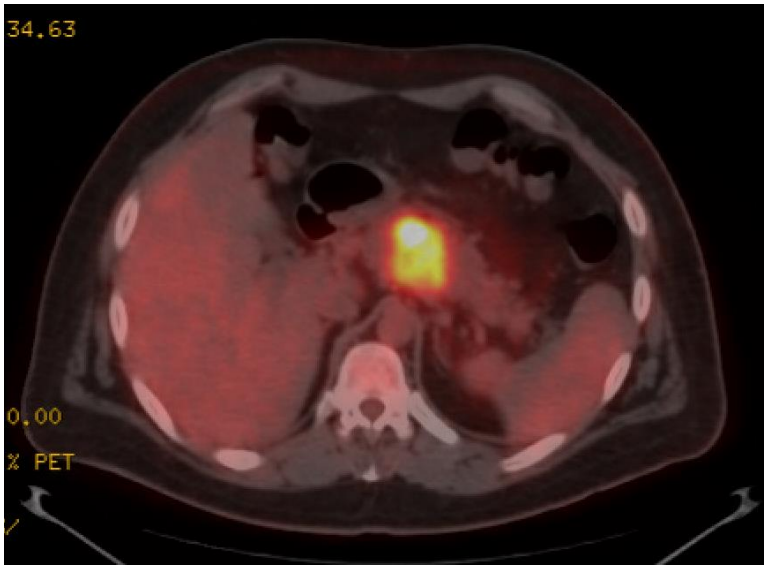

Figure 3 Axial fused image of PET/CT study shows the avid FDG-uptake in the predominantely cystic pancreatic body lesion.

3. DISCUSSION

Xanthogranulomatous pancreatitis is an extremely rare chronic destructive inflammatory process with estimated 0.74% incidence among all chronic pancreatitis in a single institution (Gaur et al., 2017). There are only a handful of reported cases in the literature. Most of these cases werefound in male patients with epigastric pain as the presenting symptoms, with mean age of 57 years old. Often these patients are misdiagnosed clinically and radiologically with pancreatic tumors, only to discover the final diagnosis of Xanthogranulomatous inflammation on post-operative biopsies. The condition is characterized microscopically by accumulation by fatopulenthistiocytes, multinucleated giant cells, plasma cells, cholesterol crevices, in addition to fibrous tissue. The hypothesized pathophysiology leading to Xanthogranulomatous pancreatitis includes a combination of (1) intracystichemorrahge, (2) stone formation, (3) mucin leakage due to tumor, and/or (4) focal duct obstruction in absence of precipitating factor (Gaur et al., 2017).

Association of Xanthogranulomatous pancreatitis with pancreatic tumors is even rarer, with only 7 reported cases in the litetrature. The most commonly associated tumors are mucinous pancreatic neoplasm, especially IPMN, with only one case reported with NET and one with mucinous cystic neoplasm (Gaur et al., 2017; Farrell, 2015; Kwon et al., 2018).

4. CONCLUSION

Our case is surprising and unique in that Xanthogranulomatous pancreatitis has been found in a male patient (which is extremely rare) with pre-existing SPEN.

Informed Consent

We reviewed the case with the ethical committee, and patient informed consent was not required for the purpose of this case study.

Ethical approval

Ethical approval was obtained from the research ethics committee of Research Advisory Council (RAC) hospital with letter number A3-3233.

Funding

This study has not received any external funding.

Conflict of Interest

The authors declare that there are no conflicts of interests.

Data and materials availability

All data associated with this study are presented in the paper.

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