Case Report

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An unusual presentation of pancreatic serous cystadenoma

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ABSTRACT

Pancreatic cystic neoplasms (PCNs) are predominantly benign entities which represent almost 50 percent of all cystic lesions of the pancreas. PCNs are often an incidental finding on abdominal imaging and are not indicated for surgical resection unless they show evidence of malignant transformation or become symptomatic due to mass effect. This report examines an unusual presentation of a PCN, in a 70 years old female with sudden onset abdominal pain, who was found to have spontaneous intraabdominal haemorrhage secondary to a benign PCN. Emergency laparotomy was performed and a distal pancreatectomy or splenectomy were required to achieve haemostasis. Incidence of spontaneous haemorrhage in a benign PCN is a rare but serious complication.

Keywords: Pancreatic cystadenoma, Pancreatic tumour, Abdominal surgery

INTRODUCTION

Pancreatic cystic neoplasms are predominantly benign entities which represent almost 50 percent of all cystic lesions of the pancreas. PCNs are often an incidental finding on abdominal imaging, and patients are usually asymptomatic. Indeed, benign PCNs are not indicated for resection unless they show evidence of malignant transformation or become symptomatic, usually due to a slowly evolving mass effect. 3.4

Here we outline the unusual case of a female in her 70s who presented with massive intraabdominal haemorrhage resulting from the rupture of a previously undiagnosed benign pancreatic serous cystadenoma.

CASE REPORT

A female in her 70's presented to an outer metropolitan emergency department with sudden onset, severe abdominal pain. The patient had a pertinent history of paroxysmal atrial fibrillation, mitral valve replacement and previous open appendicectomy. Medications included warfarin, ivabradine and flecainide.

On arrival to the ED, the patient was found to be hypotensive with a blood pressure of 50/38 mmHg which improved to 130/60 mmHg after a 1500 ml fluid bolus. In the context of flecainide, there was no cardiac compensation and her heart rate remained steady at 65bpm. The patient's abdomen was distended and guarded though not rigid. Initial bloods showed a haemoglobin of 74 and INR of 2.1.

Mobile chest X-ray demonstrated no free gas, whilst bedside focused assessment with sonography for trauma scan showed significant free fluid in bilateral upper quadrants and pelvis.

A CT mesenteric angiogram demonstrated a partly necrotic lesion involving the distal body and tail of pancreas with an extension to the splenic hilum (Figure 1 and 2). CT evidence of substantial free fluid within the

abdomen further raised concern of haemorrhage. Emergency laparotomy was immediately pursued.

Intraoperative findings were that of a large multi cystic tumour involving the body and tail of pancreas, densely adherent to the splenic hilum, left kidney and mesocolic vessels. A distal pancreatectomy and splenectomy were undertaken and haemostasis achieved. Postoperatively the patient was admitted to the intensive care unit where she remained intubated for 3 days, required four units of packed red blood cells and inotropic support. Following an extended admission for a pancreatic leak, the patient was discharged home on day 17.

Histology confirmed a serous cystadenoma without malignant features. On recent review, three years post, there was no evidence of recurrence.



Figure 1: Serous cystadenoma of the distal pancreas.



Figure 2: Serous cystadenoma of the distal pancreas.

DISCUSSION

Pancreatic cystic neoplasms are true cystic lesions of the pancreas that exist in an estimated 2-45% of the general population and account for nearly 50% of all pancreatic cysts.^{1,2}

The increasing use of cross-sectional imaging has seen a rise in the incidental diagnosis of PCNs in patients undergoing CT or MR imaging of the abdomen for unrelated indications.

These incidental lesions are largely benign, though malignant potential does exist in some subtypes. PCNs are grouped into four subtypes according to the world health organisation histologic classification.^{3,4} They are: serous cystic neoplasms (SCN), mucinous cystic neoplasms, intraductal papillary mucinous neoplasms (IPMNs): main-duct or branch-duct, and solid pseudo papillary neoplasms.

Age of presentation for PCNs is typically between the 5th and 7th decade, with the exception of the solid pseudo papillary subtype which presents earlier, between the 2nd and 3rd decade of life. Intraductal mucinous neoplasms occur equally in men and women, whilst all other subtypes have a female predominance.

Diagnosis of new PCNs is almost always incidental. Once suspected on initial imaging, of which MRI is superior, tissue diagnoses may be sought. Endoscopic ultrasound with fine needle aspiration is the method of choice.^{5,6}

A patient with a pancreatic serous cystadenoma is most likely to be asymptomatic. Symptoms, if present, usually manifest secondary to obstruction or compression of surrounding structures.

Haemorrhagic complication of PCNs is very rare, with only a handful of cases reported in the literature.⁷⁻⁹

Pancreatic serous cystic neoplasms are a wholly benign entity. Early studies suggested that serous cyst adenomas had low malignant potential however there is a growing body of evidence to support that serous cystadenomas are indeed the only PCNs that are benign. ^{10,11} It is thought that early previous reports of malignant lesions did not truly satisfy the WHO definition of serous cyst adenomas. ^{12,13}

The 2018 European evidence-based guidelines on pancreatic cystic neoplasms recommend just one year of follow-up for newly diagnosed serous cystadenomas.⁴ The rationale for this advice is the evidence that confirmed serous cystadenomas are benign and need only be operatively managed if causing compressive symptoms to adjacent organs. The rate of growth of these lesions is considered so slow that new onset of symptoms is exceedingly rare. Indeed, approximately 60% of SCNs

remain of stable size. ^{11,14-16} There is evidence to support that SCNs of the pancreatic head tend to exhibit more aggressive behaviour including faster rate of growth. ¹⁷

CONCLUSION

PCNs are common. Most are serous cystadenomas, which are benign. Serous cystadenomata may require elective surgery for mass effect. We describe a rare case where emergency surgery was required. Incidence of spontaneous haemorrhage in a benign PCN is a rare but serious complication.

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REFERENCES

- 1. Jong K, Neo CY, Hermans JJ, Dijkgraff MG, Gouma DJ, Eijck CH, et al. High prevalence of pancreatic cysts detected by screening magnetic resonance imaging examinations. Clin Gastroenterol Hepatol. 2010:8:806-11.
- 2. Chang YR, Park JK, Jang JY, Kwon W, Yoon JH, Kim SW. Incidental pancreatic cystic neoplasms in an asymptomatic healthy population. Medicine (Baltimore). 2016;95:5535.
- 3. Hruban RH, Bofetta P, Bosman FT, Carniero F, Hruban HR. Tumours of the pancreas. WHO Classification of tumours of the digestive system, 4th ed. Lyon: Int Agency for Res on Cancer. 2010: 280-330.
- 4. Pancreas TESGoCTot. European evidence-based guidelines on pancreatic cystic neoplasms. Gut. 2018;67(5):789-804.
- Sahani DV, Kambadakone A, Macari M, Takahashi N, Chari S, Fernandex CC. Diagnosis and management of cystic pancreatic lesions. AJR Am J Roentgenol. 2013;200:343-54.
- 6. Jong DK, Neo C, Mearadji B, Phoa SS, Engelbrecht MR, Dijkgraaf MG, et al. Disappointing interobserver agreement among radiologists for a classifying diagnosis of pancreatic cysts using magnetic resonance imaging. Pancreas. 2012;41:278-82.
- 7. Shinzeki M, Hori Y, Fujino Y, Matsumoto I, Toyama H, Tsujimura T, et al. Mucinous cystic neoplasm of the pancreas presenting with hemosuccus pancreaticus: Report of a case. Surg Today. 2010;40(5):470-3.

- 8. Matsumoto Y, Miyamoto H, Fukuya A, Nakamura F, Goji T, Kitamura S, et al. Hemosuccus pancreaticus caused by a mucinous cystic neoplasm of the pancreas. Clin J Gastroenterol. 2017;10:185-90
- 9. Tamura S, Yamamoto Y, Okamura Y, Sugiura T, Ito T, Ashida R, et al. A case of duodenal hemorrhage due to arteriorvenous malformation around a serous cystic neoplasm. Surg Case Rep. 2018;4:140.
- 10. Reid MS, Choi H, Memis B, Krasinskas AM, Jang KT, Akkas G, et al. Serous neoplasms of the pancreas: a clinicopathologic analysis of 193 cases and literature review with new inights on macrocystic and solid variants and critical reappraisal of so-called "serous cystadenocarcinoma". Am J Surg Pathol. 2015;39:1597-610.
- 11. Jais B, Rebours V, Malleo G, Salvia R, Fontana M, Maggino L, et al. Serous cystic neoplasm of the pancreas: a multinational study of 2622 patients under the auspices of the Internstional Association of Pancreatology and European Pancreatic Club (European Study Group on Cystic Tumours of the Pancreas). Gut. 2016;65:305-12.
- 12. Strobel O, Z'graggen K, Schmitz-Winnenthal FH, Friess H, Kappeler A, Zimmerman A, et al. Risk of malignancy in serous cystic neoplasms of the pancreas. Digestion. 2003;68:24-33.
- 13. Galanis C, Zamani A, Cameron JL, Campbell KA, Lillemoe KD, Caparrelli D, et al. Resected serous cystic neoplasms of the pancreas: a review of 158 patients with recommendations for treatment. J Gastrointest Surg. 2007;11:820-6.
- 14. Tseng JF, Warshaw AL, Sahani DV, Lauwers GY, Rattner DW, Fernandez-del Castillo C. Serous cystic neoplasms of the pancreas: tumour growth rates and recommendations for treatment. Ann Surg. 2005;242:413-9.
- 15. Das A, Wells C, Nguyen CC. Incidental cystic neoplasms of the pancreas: what is the optimal interval of imaging surveillance. Am J Gastroenterol. 2008;103:1657-62.
- 16. Palaez-Luna MC, Moctezuma VC, Hernandex CJ, Uscanga-Dominguez LF. Serous cystadenomas follow a benign and asymptomatic course and do not present a significant size change during follow up. Rev Invest Clin. 2015;67:344-9.
- 17. Khashab MA, Shin E, Amateau S, Canto MI, Hruban RH, Fishman EK, et al. Tumour size and location correlate with behavior is pancreatic serous cystic neoplasms. Am J Gastroenterol. 2011;106:15221-6.

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