Giant pancreatic incidentaloma: Report of a case and literature review

Petros Charalampoudis\textsuperscript{a, b}, Dimitrios Dimitroulis\textsuperscript{a}, Eleftherios Spartalis\textsuperscript{a}, Chrysovalantis Vergadis\textsuperscript{b}, Anastasios Stofas\textsuperscript{c}, Theodore Karatzas\textsuperscript{a}

\textsuperscript{a} 2nd Propedeutic Department of Surgery, Athens University Medical School, Laikon General Hospital, Athens Medical School, Athens, Greece
\textsuperscript{b} Department of Radiology, Laiko General Hospital, Athens, Greece
\textsuperscript{c} Department of Pathology, Athens University Medical School, Laiko General Hospital, Athens, Greece

\textbf{A R T I C L E   I N F O}

Article history:
Received 23 January 2012
Received in revised form 2 April 2012
Accepted 17 April 2012
Available online 24 April 2012

Keywords:
Pancreas
Incidentaloma
Serous cystadenoma

\textbf{A B S T R A C T}

\textbf{INTRODUCTION:} Asymptomatic lesions of the pancreas, referred to as ‘incidentalomas’, have appeared with increased frequency in recent years. Giant incidentalomas have rarely been reported in the literature.

\textbf{PRESENTATION OF CASE:} We report herein a rare case of a giant cystic pancreatic incidentaloma measuring 12.7 cm $\times$ 8 cm, which was found in an otherwise healthy male patient during a routine genitourinary imaging work-up. The patient underwent a distal pancreatectomy and splenectomy; the pathology report demonstrated a giant serous cystadenoma of the body and tail of the pancreas.

\textbf{DISCUSSION:} The management of pancreatic incidentalomas is challenging. While solid lesions almost always warrant surgery, there is ongoing debate concerning the management of cystic lesions that are found incidentally in the pancreas and have no clinical manifestations.

\textbf{CONCLUSION:} We report herein an interesting case of a voluminous incidental cystic pancreatic lesion. The appropriate approach and the decision whether to operate or not in such cases can be puzzling to the physician.

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1. \textbf{Introduction}

Continuous improvement in imaging resolution during the past years has led to increasing reports of incidental lesions. Although the most prevalent site of such lesions is the adrenal gland, incidentalomas can also be located on other organs, such as the kidney, pituitary, parotid gland, liver, heart, prostate and pancreas. Like their symptomatic counterparts, incidental lesions can be solid or cystic. While solid pancreatic masses, even without any clinical manifestations, call for surgical management, there is considerable debate regarding the management of cystic asymptomatic lesions found incidentally in the pancreas. We report herein a case of an otherwise healthy male individual who was admitted to our Department after a giant pancreatic lesion was found incidentally during a routine genitourinary ultrasound. We also review the literature concerning the ongoing debate on when and whether to perform surgery for large, cystic pancreatic lesions.

2. \textbf{Case report}

A 74-year-old male was admitted to our Surgical Department for management of a giant asymptomatic pancreatic mass, identified in a routine genitourinary ultrasound. The lesion was hypoechoic, well defined, and occupied the body and tail of the pancreas, without any other abnormal findings. The patient did not mention any abdominal discomfort or other complaints. He denied any alcohol consumption and his past medical history was otherwise unremarkable. Physical examination of the abdomen was negative for any palpable mass. Routine serum chemistry profile ranged within normal levels. His serum CA 19-9, α-fetoprotein and CEA tests were normal. CT scan of the abdomen revealed a giant tumour measuring 12.7 cm $\times$ 8 cm that was localized to the body and tail of the pancreas. The lesion was non-homogeneous and hypodense compared to normal pancreatic tissue and demonstrated a cystic component with multiple septa. There was no dilation of the main pancreatic duct and no other abnormality such as a hepatic mass or lymphadenopathy was detected (Fig. 1a). Abdominal MRI showed a giant pancreatic lesion composed of numerous small cysts with fibrous septa between them and a fibrous central scar that was enhanced on delayed contrast MR sequences. No visible communication between the cysts and the pancreatic duct was noted (Fig. 1b and c).

After a thorough preoperative workout, an exploratory laparotomy confirmed the presence of an enormous, well-circumscribed, more solid-appearing lesion, occupying the body and tail of the pancreas. The lesion was attached to the splenic porta and the transverse mesocolon, without causing any mesocolon vasculature compromise. The patient underwent an en-block distal pancreatectomy and splenectomy under general endotracheal anaesthesia (Fig. 2a and b). His postoperative course was uneventful, and he was discharged on the 7th postoperative day.

\textsuperscript{*} Corresponding author.
E-mail address: pcharalampoudis.laiko@gmail.com (P. Charalampoudis).

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http://dx.doi.org/10.1016/j.ijscr.2012.04.009
Fig. 1. (a) Transverse contrast enhanced CT obtained during pancreatic phase shows a well-circumscribed encapsulated low-attenuation inhomogeneous tumour of pancreatic body and tail with internal septa. (b) MRI T2-weighed coronal image demonstrates very high intensity of the mass with polycystic pattern and multiple fibrous septa. (c) MRI T1-weighed gadolinium enhance delayed image depicts enhancement of internal septa and central scar (arrow shows the scar tissue area).

The pathology report revealed a pure serous pancreatic cystadenoma. The tumour was grossly well circumscribed. On cross-section, it was sponge-like and composed of numerous tiny cysts filled with serous fluid. The cysts were arranged around a central fibrous stellate scar. Histologically, the cysts ranged from 0.01 to 0.5 cm and were lined by a single layer of cuboidal epithelial cells. Their cytoplasm was almost clear, and their nuclei were centrally located, round, uniform and hyperchromatic. Occasionally, the neoplastic cells formed intracystic, micro-papillary projections. The periodic acid-Schiff (PAS) stain was positive, whereas PAS-diastase was negative due to the presence of abundant intracytoplasmic glycogen. The neoplastic cells were immunoreactive for epithelial membrane antigen and cytokeratins 7, 8/18 and 19 owing to the epithelial nature of this neoplasm (Fig. 3a and b).

3. Discussion

Incidentally discovered solid organ lesions that cause no symptoms (incidentalomas) are increasingly being detected due to emerging imaging resolution during the past years. Common incidentalomas have been described in the liver, kidney and adrenal glands.1 There has also been an increase in the occurrence of unusual, asymptomatic tumours of the pancreas. The first report of pancreatic incidentaloma in the literature was published by Kostiuk in 2001 (2 cases) and several series have since been reported.2 Such incidental pancreatic tumours include serous cystadenomas, mucinous cystadenomas, mucinous cystadenocarcinomas, nonfunctional neuroendocrine tumours, papillary cystic and solid tumours, and intraductal papillary mucinous neoplasms (IPMN).1
Mucinous cystic tumours, serous cystadenomas, and intrapapillary mucinous neoplasia (IPMN) comprise more than 90% of overall primary cystic neoplasms of the pancreas. Cystic islet cell tumours, solid pseudopapillary tumours, and other pathologies with cystic appearance are less common.

The management of patients with these findings in organs, such as thyroid, adrenal gland, and lung, has been well described, and treatment algorithms have been proposed. However, literature on pancreatic incidentalomas is lacking, and given their increased incidence, guidelines regarding their management are yet to be developed.

Multislice contrast-enhanced CT scan is an excellent examination for the initial detection of pancreatic lesions and for characterization through visualization of calcifications, septa, nodules and findings suggestive of pancreatitis. Magnetic resonance imaging (with a combination of rapid T2-weighted sequences and unenhanced and contrast enhanced T1-weighted sequences) has the added advantage of providing better tissue characterization, allowing for the optimal evaluation of the internal architecture of a cyst and optimal demonstration of enhancing soft-tissue element. In the future, newer techniques including F-18-fluorodeoxyglucose positron emission tomography (FDG-PET) may help in distinguishing benign and malignant pancreatic incidentalomas.

To date, the existing methods to characterize pancreatic cysts lack the accuracy to distinguish between low-risk and high-risk lesions. The assessment of the cysts based purely on morphologic features cannot accurately distinguish the type of cyst or predict malignant transformation. Mucinous tumours or macrocystic adenomas may be either cystadenomas or cystadenocarcinomas. Mucinous cystadenoma is not a malignant lesion but has the potential for malignant transformation with rapid progression and should thus be viewed as premalignant or potentially malignant. Surgical resection is recommended in patients with appropriate surgical risk. Cystadenocarcinoma is a malignant tumour and should be treated with aggressive surgical intervention. IPMN is a newly described pancreatic neoplasm defined by the World Health Organization in 1996. This lesion represents a premalignant or frankly malignant condition with a histologic spectrum ranging from adenoma to infiltrating carcinoma. The proportion of tumours that are malignant varies from 21% to 37%. Surgical resection is recommended in all appropriate-risk patients. Recent findings imply that many incidental pancreatic cysts are small IPMN-branched duct lesions (IPMN-Br), which have a low potential for malignancy. Fernandez-del Castillo pointed out that IPMN was the most frequent type of symptomatic and asymptomatic cysts and Levy reported that 39% of IPMNs are incidental, 50% of IPMNs are IPMN-Br, and the risk of developing high-grade dysplasia or invasive carcinoma within 5 years is 63% for main pancreatic duct IPMN and 15% for IPMN-Br.

Lahav suggested that the natural course of incidental pancreatic cysts usually is benign after a median follow-up period of 48 months. However, one should be aware that mucinous cystic neoplasm is a pathologic diagnosis and it is not clear how many of our un-operated cysts were mucinous. Data from the Lahav series fails to agree with other studies that have reported rates of 20–42% of premalignant or malignant lesions in pancreatic cysts. Morphologic features derived from EUS allow the endoscopist to suggest a diagnosis in about two-thirds of cases. In a series by Ferrone et al., this fact matched the final histopathological diagnosis in 52% of asymptomatic cysts, respectively. Cytologic examination of the cyst contents often is non-diagnostic because of the inadequacy of the specimen acquired by the aspiration. The yield and sensitivity of aspirate analysis are far from satisfactory, and in many cases they do not allow for a final characterization of the cyst.

Patients with incidentally discovered pancreatic lesions and thought to have a low risk of cancer may be managed by serial observation; for example, cystic tumours <2 cm in patients with no clinical or radiologic features of malignancy. In a series by Bruzoni et al., 80% of patients who had tumours >2 cm had a malignancy, indicating that these patients are more likely to need resection, whereas those with smaller tumours may simply be monitored. The Massachusetts General Hospital Group published an algorithm in 2003 to manage patients with incidental pancreatic cysts. According to their algorithm patients with cysts <2 cm or those with cysts >2 cm combined with negative carcinoembryonic antigen (CEA), mucin, or cytology levels can be observed clinically. The International Consensus Guidelines for Management of Intraductal Papillary Mucinous Neoplasms and Mucinous Cystic Neoplasms of the Pancreas concluded that patients with cystic lesions ≥3 cm, or high malignancy-risk patient with lesions between 1 and 3 cm, would benefit from resection.

In our case, the giant pancreatic lesion described herein turned out to be a serous cystadenoma. Although there is no management consensus in the pertinent literature regarding large asymptomatic pancreatic cysts, several authors suggest that such lesions mandate close observation provided that preoperative diagnosis precludes mucinous cystic neoplasms or cystadenocarcinoma. However, Strobels suggests that serous cystic neoplasms of the pancreas do have malignant potential with a risk of malignancy of 3% and should be surgically treated if the operative risk is acceptable. In cases of non-definitive diagnosis of incidental pancreatic cysts, given the safety of pancreatic resection in specialized centers, Lahat et al. support resection as the treatment of choice.
The generally benign course of serous cystic neoplasias has led to an ‘observational’ trend for many years; however, this policy has recently been debated. The decrease in perioperative mortality after major pancreatectomy during the last two decades probably accounts in part for the altered consensus regarding a more aggressive approach with resection.  

In our case, the lesion was very large, measuring 12.7 cm × 8 cm. There appears to be a current trend towards surgery for serous cystic lesions with a size >4 cm. Any presence of symptomatology or diagnostic uncertainty equally warrants surgery. In our case presented herein, apart from the large size of the cyst, imaging failed to exclude serous malignancy. Upon diagnostic doubt, some authors recommended cyst aspiration with fluid analysis; more recently, endoscopic ultrasound (EUS) with fine-needle aspiration (FNA) has been suggested as a method to differentiate between benign, premalignant, and malignant lesions. However, aspiration either percutaneously or endoscopically via EUS has significant spillage potential and is therefore not routinely recommended.  

Due to the large size of the cyst, spillage risk was significant in case an FNA was performed. Our patient was a 72 year old, otherwise healthy individual. Spinelli et al. mandate surgical excision for pancreatic cysts that increase under observation, manifest with symptoms or are discovered in healthy, older patients. He also found that older patients over 70 years are more likely (p < 0.02) to have premalignant or malignant cystic pancreatic neoplasms. Serious cystadenoma of the pancreas presents at an earlier age (61 years) than serious cystadenocarcinoma (66 years; p = 0.056) and are symptomatic in the majority of patients.

4. Conclusion

A thorough understanding of the current trend towards asymptomatic cystic lesions in the pancreas along with weighing of individual patient factors are essential to guide a surgical team towards the optimal management of such cystic pancreatic tumours. In this interesting case, the large size and doubtful preoperative diagnosis combined with a modern, literature-based aggressive surgical practice for this type of lesions, led us to opt for surgery in this, otherwise healthy, 72 year old patient.

Conflicts of interest

The authors declare that there is no conflict of interest.

Funding

None.

Ethical approval

Written informed consent has been obtained by the patient.

Author contributions

Petros Charalampoudis and Dimitrios Dimitroulis equally contributed to the writing of this paper. Petros Charalampoudis, Eleftherios D. Spartalis, Dimitrios Dimitroulis and Theodore Karatzas performed the operation. C. Vergadis analyzed the imaging studies. A. Stofas conducted the pathology report of the specimen.

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