Giant serous microcystic pancreas adenoma

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Abstract

Serous cystadenomas are rare tumors comprising 1-2% of exocrine pancreas tumors. They are mostly known as benign conditions but malign transformation as serous cystadenocarcinoma is also reported. It is usually seen in females. Non-specific symptoms, such as abdominal pain or symptoms due to mass affect, are usually seen. A 64-year old female patient was investigated for abdominal pain. Physical and laboratory findings were normal. Abdomen ultrasonography confirmed an 11×9.5 cm solid cystic lesion and abdomen computed tomography scan confirmed a 12×11 cm lobulated cystic solid lesion which had central cystic necrotic areas extending from liver hilus inferiorly. Fine needle biopsy confirmed benign cytology and trucut biopsy of the pancreatic mass reported chronic inflamation. Nevertheless, this mass could have malignant contents and transformation potential. A laparatomy was decided due to patient's symptoms and mass effect. Due to vascular invasion of the tumor, Whipple procedure was performed. The pathology report confirmed serous microcystic adenoma. These rare tumors are usually benign but pre-operative malignity criterias are not identified. There are few differential diagnostic tools for excluding malignity. We suggest surgical resection as best treatment approach for selected cases.

Introduction

Serous cystadenomas are rare tumors comprising 1-2% of exocrine pancreas tumours and 1% of all pancreas tumors.[1,2] They are mostly known as benign conditions but malign transformation as serous cystadenocarcinoma has also been reported.[3-8] Cystic neoplasms are divided into four groups; serous cystadenoma, musinous cystadenoma, intraductal papillary musinous tumor, and rare cytstic neoplasm.[9] According to World Health Organization classification there are microcystic and oligocystic variants, the microcytic form being more common.[10] It is usually seen in females in the sixth decade of life and is mostly asytmptomatic.[11] Non-specific symptoms, such as abdominal pain, are more common, but symptoms resulting from mass pressure are also not unusual.[12-14] Fifty percent of cases have lesions located on the head of the pancreas.[15] Several studies have reported tumors ranging in size from 1-2 cm to 25 cm.[16]

In this study, we report an unusual symptomatic giant serous microcystic adenoma and its diagnosis, malignant criteria and treatment approaches are examined along with findings described in the literature.

Case Report

A 64-year old female patient with abdominal pain came to us for a consultancy. She had had type 2 diabetes mellitus for 18 years and had had a cholecystectomy 15 years previously. She had been examined ten years ago for her abdominal pain and a pancreas mass was detected in another city hospital. Pancreas fine needle biopsy confirmed benign cytology; she underwent no further check-ups. Three years ago, abdominal pain and nausea increased and she underwent trucut biopsy from pancreatic mass in another center. The biopsy report confirmed chronic inflamation. Furthermore, abdomen ultrasonography (USG) confirmed an 11×9.5 cm solid cystic lesion located from the left lobe inferior segment of liver extending to pancreas and bursa omentalis. Abdomen computed tomography (CT) scan confirmed a 12×11 cm lobulated cystic solid lesion which had central cystic necrotic areas extending from liver hilus inferiorly (Figure 1). This mass mobilated the right and left portal vein, inferior vena cava, left portal vein and superior mesenteric artery. Its pelvic border above right kidney and soft tissue was not clear. Given these radiological findings she was hospitalized in our unit. During hospitalization, physical examination revealed nothing of significance and her vital signs were normal. She had mild elaveted liver enzymes: alanine minotransferase 48 mg/dL (normal range 0-40); aspartate aminotransferase 128 mg/dL (normal range 0-42). Her blood glucose levels were regulated by high-dose insuline (80 units daily). Other blood and urine tests and cancer biomarkers were in the normal range.She had undergone another trucut biopsy from pancreatic mass; the biopsy report confirmed benign mesothelial cystic lesion. Nevertheless, since this mass could have malignant contents, upper and lower gastrointestinal endoscopies (GIS) were performed. Upper GIS examination revealed antral hyperemic and nodular areas, and a duodenal noduler lesion. Owing to mass effect, bulging to anterior duodenal tissue was revealed. Multiple biopsies were taken and results were reported as normal tissue. Neither did colonoscopy show any remarkable finding. It was decided to perform a laparatomy because of the patient's symptoms, malignity potential and mass effect. During exploration, pancreatic mass was seen in a wide area but there was no invasion to the borders. Due to vascular invasion of the tumor, Whipple procedure was performed (Figure 2). The patient was followed in the internal care unit until postoperative day 3 and discharged postoperative day 12 without any complications. The pathology report confirmed a tumor 15.5 cm in diameter. This report confirmed serous microcystic adenoma (Figures 3,4). There was no organ invasion or metastatic lymph tissue.

Discussion

Serous cystadenomas are rare benign tumors of the pancreas.[17] There are also some reported cases of malignant transformation as serous cystadenocarcinoma.[3-8] Serous cystadenomas comprise 25% of pancreas cystic neoplasms and 1% of all pancreas tumours.[2,17] It is usually seen in females in the sixth decade of life (our patient was also 64 years of age) and is mostly asytmptomatic.[11] Most of the symp-

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toms are non-specific, such as abdominal pain, nausea, vomiting, and weight loss. One-third of the cases are asymptomatic and it is diagnosed incidentally.[17] The Compagne and Oertel series reported an average tumor size of 10.8 cm and 29% asymptomatic patients. Pyke *et al.* reported 33% asymtomatic cases.[18] Our patient also had non-specific symptoms and tumor size was 15.5 cm.

There are many radiological imaging techniques that are useful, such as CT, magnetic resonance imaging, endoscopic retrograde cholangiopancreatography, magnetic resonance cholangiopancreatography, and endoscopic ultrasonography. The lesion is usually located on the head of the pancreas,[15] as in our patient. Serous cystadenomas are formed from multiple cysts which are seperating from each other with thin septas; it looks like honeycomb. USG shows at least 6 loculated cysts each less than 2 cm in size.[19] Abdomen CT shows calcifications with a central scar resembling sun explosions which are patognomonic but only seen in 30-40% of cases.[20] Despite the advantages of all these imaging techniques, preoperative diagnosis rates are only 40%.[21] Serous cystadenomas can also be seen with other pancreatic and non-pancreatic neoplasms.[22-24] Our patient did not have any other neoplasm. There is no consensus on diagnostic and treatment approaches but pre-operative diagnosis, safe resection and conservative treatment are changing the treatment strategy.[21] Symptomatic cases usually undergo surgery. Drainage procedures are not suitable for these tumors. Tumors located on corpus and tail of the pancreas distal pancreatectomy is a suitable resection; however, for tumors located on the head of the pancreas the whipple procedure represents a better approach.[21] In our case, we used a whipple procedure for the symptomatic tumor located on the head of the pancreas. Some authors suggest conservative treatment for asymptomatic and high-risk patients.[10,25]

However, conservative treatments have some disadvantages, such as the unknown nature of the tumor (as in our case) and the probability of complications.[26]

Serous microcystic adenoma, also known as glycogen rich cystadenoma, is composed of microglandular cysts lined by clear epithelial cells histologically rich in glygogen, which were seperated by fibrocollagenous stroma. The expression of keratin in clear epithelial cells resembled that in ductal or centroacinar cells, but not that in acinar cells.[27] Serous microcystic adenomas have a characteristic spongy appearance, but this varies in some reports. Cells are cuboidal in shape and arranged in a single layer that is usually flat but may become pleated in a collapsed cyst forming small papillae projecting into the cyst lumen.[27]

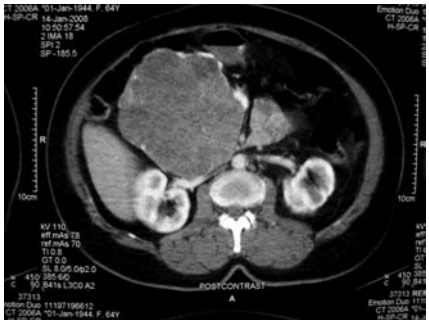

Figure 1. Abdominal computed tomography section showing a 12×11 cm giant cystic lobulated solid mass located on the head of the pancreas.

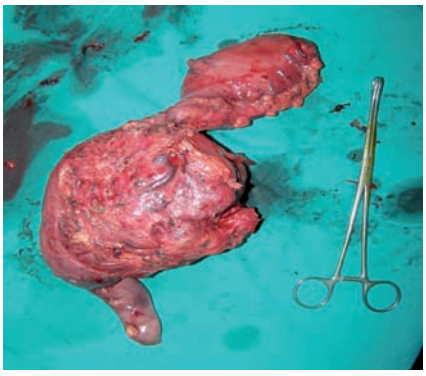

Figure 2. Macroscopic appearance of the mass.

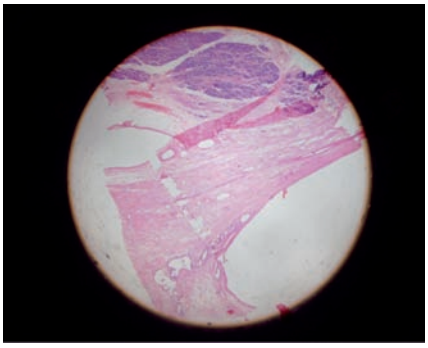

Figure 3. Cystic tumoral pathology near the non-neoplastic pancreatic tissue (Haematoxylin and Eosin 12.5×).

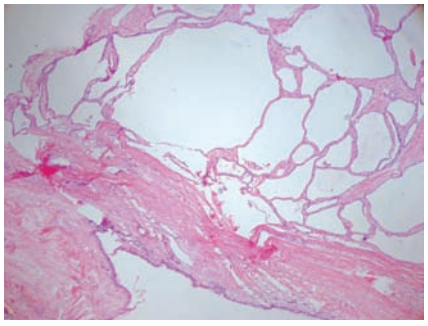

Figure 4. Serous microcystic adenoma. Multiple epithelial cysts of various dimensions are separated by fibrocollagenous septae (Haematoxylin and Eosin 100×).

Serous microcystic adenomas are frequently associated with diabetes mellitus (24-36% of cases), as in our case. The association with diabetes mellitus is due to islet cell damage caused by the tumor or may be coincidental to patient age.[27] The tumor occurs sporadically or as a part of von Hippel-Lindau disease that has a much wider range of tumor number, size and distribution within the pancreas.[27]

Conclusions

In conclusion, serous microcystic pancreas neoplasms are rare tumors. They are usually benign but pre-operative malignity criteria have not been identified. The lack of effective tools for differential diagnosis from other cystic malignant neoplasms leads us to suggest resection as being the best treatment approach in selected cases.

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