

MACROCYSTIC VARIANT OF SEROUS CYSTADENOMA PANCREAS IN A CASE OF DORSAL BUD AGENESIS OF PANCREAS-A RARE CASE REPORT

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ABSTRACT

Macrocytic variant of serous cystadenoma (MSC) of pancreas is an unusual benign tumor seen only 10% case of serous cystadenoma. Ages of reported cases are more than 60 years. Herein, we report a case of 45 year old female diabetic patient presents with upper abdominal pain. On CT image, cystic lesion having central popcorn calcification with complete agenesis of neck, body, tail of pancreas is noted. Initially it was thought to be pancreatic pseudocyst in dorsal agenesis of pancreas on upper GI endoscopy. Later the patient undergone 3T MRI, biochemical analysis of aspirated fluid and trucut biopsy followed by excisin biopsy and proven to be macrocytic varint of serous cystadenoma in case of dorsal agenesis of pancreas.

Introduction

Serous cystadenoma of the pancreas is a bening cystic epithelial neoplasm composed of serous-type, glycogen-rich epithelial cells. It is an uncommon and essentially benign pancreatic tumor. The majority of serous cystadenomas are histologically microcystic. The macrocytic subtype of serous cystadenoma has been reported only on rare occasions. Prevalence of macrocytic variety is 10% of all serous cyst adenoma. The radiologic features of a macrocytic serous cystadenoma (MSC) may resemble a pseudocyst. Thus, it is often difficult to make a proper diagnosis. MSC usually occurs in patients over 60 years preoperatively, with a mean age of 54 years. Herein, we report on an MSC in a 45-year-old female patient, with dorsal agenesis of pancreas. To our knowledge, our patient is the younger and unique to present with such tumor associated with dorsal bud agenesis in the literature.

Case Report

A 45-year-old diabetic female was admitted to our institution in september 2014 with history of recurrent upper abdominal discomfort. Abdominal ultrasonography and computed tomography were performed to investigate his complaint of recurrent upper abdominal discomfort. This led to the discovery of a cystic lesion with central coarse calcification in the head of pancreas with complete agenesis of neck, body, tail of pancreas. He had experienced intermittent upper abdominal pain after that. The patient had no history of alcohol consumption, abdominal trauma, or pancreatitis. The physical examination was unremarkable. Laboratory data, including hematology, general biochemistry, and urinalysis, were within normal limits except high blood sugar level. she was undergone trucut biopsy but the report was inconclusive due to inadequacy of tissue sample. Aspirated

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fluid from cystic lesion shows low values of CEA (0.8 ng/ml) and amylase (12.24 iu/l). Abdominal ultrasonography showed a pancreatic cystic lesion with central calcification at pancreatic head measuring 4.2 cm in diameter. An abdominal CT scan showed a well defined hypodense cystic lesion of 4.5 x 3.1 x 3.0 cm with central popcorn calcification in pancreatic head whereas rest of the pancreas was agenetic. EUS shows multiloculated cyst with parenchymal calcification, reported as CCP with pseudocyst. While the clinical diagnosis was a pancreatic pseudocyst as suggested by the abdominal CT and EUS, a mucinous neoplasm with a definite malignant potential could not be ruled out. On 3T MRI, a multiloculated fluid intensity cyst with central coarse calcification measuring 48 x 30 mm is noted at pancreatic head with complete dorsal pancreatic agenesis including duct of santorini. Largest cyst measures 28 x 24 mm. With such features our diagnosis was more in favour of macrocystic serous cystadenoma. Thereafter, as the patient was complaining of intractable abdominal pain, he underwent an exploratory laparotomy with pancreatic cystectomy with the intent to establish a pathologic diagnosis. Macroscopically, there was a well-defined, multilocular cyst in the pancreas, which was filled with a clear watery fluid with central calcified area, no solid part identified. Microscopically, the cystic wall lining was composed of simple cuboidal or flattened epithelial cells with clear cytoplasm. The nuclei were rounded to oval in shape, uniform, and centrally located. Periodic acid-Schiff (PAS) staining was focally positive in the cytoplasm of the epithelium, and was negative after diastase digestion. Mucin staining was negative. Therefore, the tumor was diag-

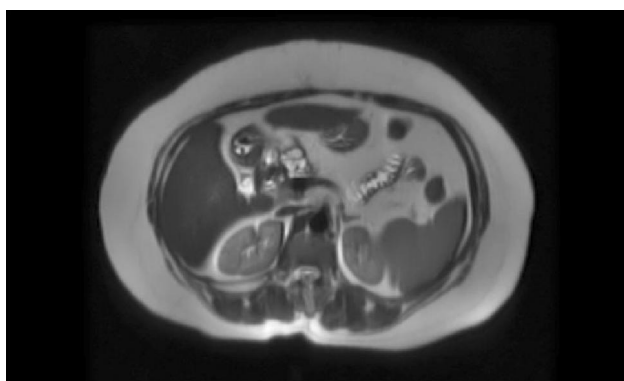


Figure 1: Axial T2 scan show multiloculated fluid intensity cystic lesion with central coarse calcification at pancreatic head with dorsal agenesis.

nosed to be a macrocystic serous cystadenoma of the pancreas. The postoperative course was uneventful. The patient was well for 1 month after surgery and is currently being followed up through our out patient department.

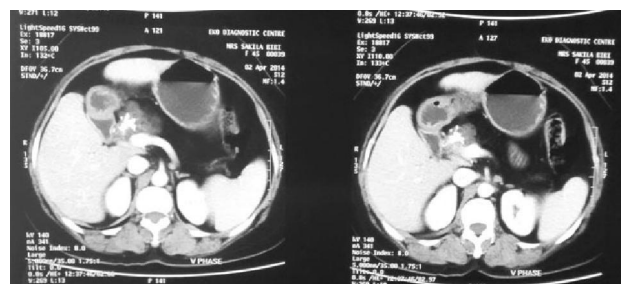


Figure 2: Axial CECT show hypodense cystic lesion with central popcorn calcification at pancreatic head in dorsal agenesis of pancreas.

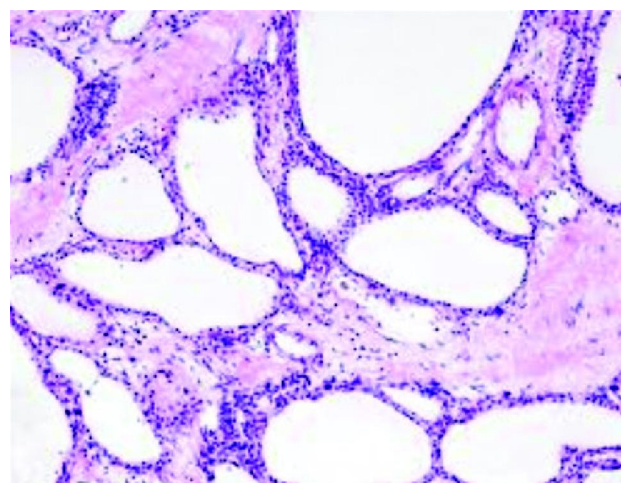


Figure 3: Cystic wall lining was composed of simple cuboidal or flattened epithelial cells with clear cytoplasm and the nuclei were rounded to oval in shape, uniform, and centrally located

Discussion

The macroscopic features of a macrocystic serous cystadenoma can be unilocular or oligocystic with each cyst having a diameter of 2.5-8 cm. Our case presented as multilocular cyst largest having 25 mm diameter. This entity was first reported by Lewandrowski et al. However macrocystic serous cyst adenoma with dorsal agenesis of pancreas is a rare case ever documented in literature. The radiological features of MSC may resemble those of a pseudocyst or a mucinous cystadenoma; the latter has the risk for malignant progression, whereas the prognosis for an MSC is much better. Therefore, it is important

to make a correct diagnosis before surgery so that appropriate surgical management can be followed. However, it seems difficult to make the distinction by imaging studies preoperatively, and almost all cases are diagnosed after surgery by histologic examination. Procacci et al. reviewed 30 cases of serous cystadenoma of the pancreas based on their imaging findings and concluded that it was impossible to diagnose macrocystic serous cystadenoma correctly by preoperative imaging. Lewandrowski et al. also found that the radiological features of their 5 cases of macrocystic serouscystadenoma were indistinguishable from those of mucinous cystic neoplasms, and when unilocular, could be confused with pseudocysts. Nguyen et al. encountered a case of an MSC of the pancreas, and they made a correct diagnosis using preoperative percutaneous needle aspiration.

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