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Case report

Mature cystic teratoma of the pancreas: Role of endoscopic ultrasound

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ABSTRACT

Cystic teratomas are a rare type of germ cell neoplasms derived from one or more germ layers. They can be classified as mature and immature teratomas based on the maturity of neuroectodermal elements within the tumor. Mature teratomas are benign, well-differentiated lesions, which may be solid or cystic. Mature cystic teratomas (MCTs) are also called dermoid cysts – a term likely coming from early surgical literature when the resected cysts resembled skin. Immature teratomas are malignant, undifferentiated tumors and generally solid. The most common location for MCTs is sacrococcygeus followed by ovaries and testes. MCTs in the pancreas are extremely rare with only few published case reports. Diagnosis of teratomas is challenging since there are no definitive preoperative diagnostic tests or pathognomonic findings. Even though EUS is commonly used for diagnosis of pancreatic cysts, the imaging features and cyst fluid characteristics of MCTs on EUS FNA have not been described. We describe a challenging case of a 65-year-old patient who had a 9 cm × 7 cm MCT in the body of the pancreas and discuss the role of EUS in diagnosis and management of these rare lesions. Complete surgical removal seems to be the current standard of care.

Case presentation

A 65-year old man with no significant medical history presented with occasional abdominal pain and early satiety for the last few months. Computerized tomography (CT) scan showed an 8 cm unilocular cyst in the body of the pancreas compressing the stomach (Fig. 1). The visualized pancreatic parenchyma appeared otherwise normal. The patient denied excessive alcohol use, abdominal trauma or abdominal surgery. There was no history to suggest prior episodes of acute pancreatitis. The patient underwent endoscopic ultrasound (EUS), which showed a 9 cm × 7 cm unilocular, well-limited, anechoic cyst occupying the entire body and tail of the pancreas. There was some debris within the cyst (Fig. 2). Aspiration of cyst fluid revealed thin, brownish aspirate. The cyst fluid carcinoembryonic antigen (CEA) concentration was 7083 ng/ml and the amylase level 221 U/ml. Cytological examination showed macrophages, few mixed leukocytes and absence of mucin. Due to persistence of symptoms and patient's reluctance to undergo surgical evaluation, an EUS guided transgastric cyst-gastrostomy was performed using standard technique. One 10Fr, 4 cm double pig-tailed plastic biliary stent was placed from the stomach into the cyst (Fig. 3). About 200cc of thin, brownish fluid immediately flowed out after placement of stent. There were no immediate complications and patient was discharged home the same day. However, 2 days later he developed right shoulder pain without abdominal localization. He did not have shortness of breath, fever or chills and was tolerating regular diet. The pain did not respond to acetaminophen treatment and he was evaluated in the hospital. Physical examination was pertinent for stable vital signs, normal breath sounds, soft, non-tender but distended, tympanic abdomen. A CT scan showed that the cyst-gastrostomy stent was in satisfactory position. The cystic lesion had slightly decreased in size measuring 8 cm × 7 cm.

The patient was managed conservatively with antibiotics, pain medications and was discharged. Antibiotics and pain medications were not continued at discharge. A follow up CT of the abdomen 4 weeks later showed complete resolution of the pneumoperitoneum. After further discussions, patient agreed to have surgical evaluation. A spleen preserving distal pancreatectomy was

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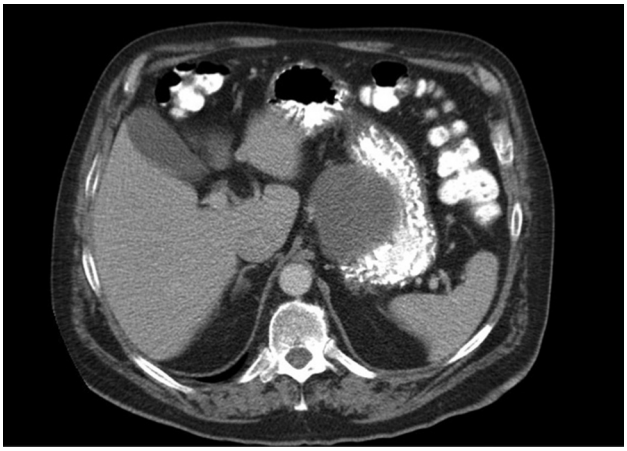

Fig. 1. Computerized tomography (CT) scan showed a 8 cm unilocular cyst in the body of the pancreas compressing the stomach.

performed. The pathology examination showed a unilocular cystic lesions measuring 7 × 7 × 3.5 cm, mainly lined by mature squamous epithelium with surrounding collections of mature lymphocytes and lymphoid follicles. Other types of epithelium were also observed in the wall of the lesion including ciliated respiratory-type epithelium and intestinal type epithelium with foci of goblet cells (Fig. 4). No malignant transformation or immature tissues were observed. The overall histology features were consistent with a benign mature cystic teratoma (dermoid cyst). The remaining pancreas was unremarkable.

Discussion

The pancreas is a very rare primary site for MCTs. MCTs are believed to arise during embryogenesis when migrating germ cells become "misplaced" en route to their appropriate organs [1]. MCTs can, therefore, be lined by any of the three germinal layers - ectoderm, endoderm, and mesoderm. To our knowledge, only 36 cases of pancreatic MCTs have been reported in English language publications [11].

MCTs in the pancreas are more common in men (59% men, 41% women) [1]. The reported age at diagnosis varies between 4 months and 75 years (mean 36 years). MCTs can develop anywhere in the pancreas but are more commonly described in the body

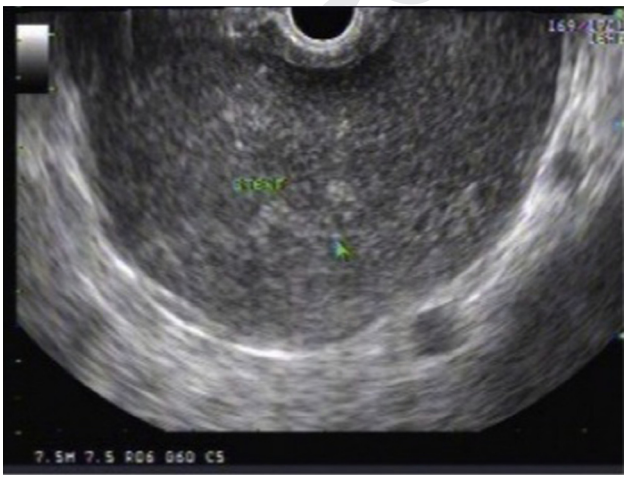

Fig. 2. Endoscopic ultrasound image of the pancreatic cyst.

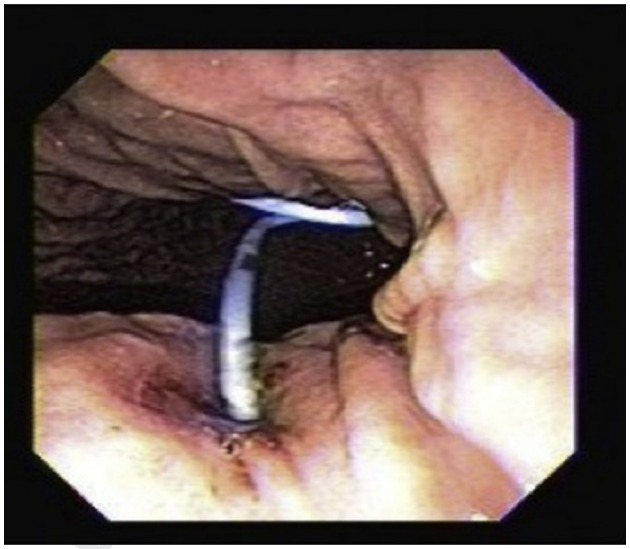

Fig. 3. Cyst gastrostomy with placement of pig-tailed stent.

(47%) or the head (41%). Their size ranges from 2 to 22 cm with the average size being approximately 7 cm. As compared to the adult population, we found only 8 published cases of MCTs in children. The cyst sizes in these cases ranged from 9 to 18 cm, with majority located in the body of the pancreas [17]. No extrapancreatic MCTs or other tumors were noted in these 8 cases. While most patients with pancreatic MCTs are asymptomatic and lesions are discovered incidentally, patients may complain of nonspecific gastrointestinal symptoms such as nausea and vomiting, anorexia, fatigue, abdominal pain and back pain [5,6]. In the pediatric age group vomiting is the most common feature. Rarely, patients may have an acute presentation as related to spontaneous rupture, however not reported in pancreatic MCTs [14]. Choi et al. reported no significant difference between ruptured and unruptured teratomas in terms of wall thickness, location of mass and tumor size [14]. Palpable abdominal mass and/or abdominal tenderness are the most common physical exam findings in all age groups. Since the signs and symptoms are non-specific, the diagnosis of MCT is made on cross sectional imaging such as abdominal ultrasound, CT or magnetic resonance imaging (MRI). The radiologic appearance of pancreatic MCTs depends on their composition [2]. Presence of fat, fat-fluid levels, and calcium within a cyst lumen are considered to be highly suggestive of a mature teratoma, but these are seldom seen. The imaging findings for MCTs in the published case reports have been fairly non-specific and the findings had sufficient overlap with other pancreatic cysts such as pseudocyst, serous or mucinous cystadenomas or solid pseudopapillary tumors [1]. Hence diagnosis of MCT remains difficult. Our patient had a fairly typical presentation in terms of age, sex and presenting complaints. Imaging did not show typical features and it was reported as a cyst with a broad differential.

When a definitive diagnosis of a pancreatic cyst type remains in doubt, the next step is surgery or EUS guided fine needle aspiration (FNA) [18]. Some advocate a surgical approach for symptomatic cysts or cysts >3 cm with the notion that larger cysts are more likely to be premalignant or malignant and surgery may relieve the symptoms [5,6,18]. However, cysts >3 cm may be serous cystadenomas or other benign tumors making surgery unnecessary. Cysts in the head of the pancreas often may require more extensive surgery such as a Whipple type surgery, which may result in more clinical post-surgical complaints. Hence, for indeterminate cysts with no imaging features of malignancy, attempt to get more