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CASE REPORT

Robot Pancreatic Resection for a Mature Cystic Teratoma of the Pancreas – A Case Report and Review of Literature

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ABSTRACT

Dermoid cysts of the pancreas are rare. A Seventy-five-year-old lady was evaluated for epigastric pain. A CT scan of the abdomen revealed a lesion in the pancreatic tail measuring 2.9×3.1 cm. She had normal tumour markers. An Endoscopic Ultrasound-Guided Fine Needle Aspiration revealed a 20 mm solid mass and pus was aspirated. The cytology was inconclusive and the fluid too viscous for biochemical tests. An interval scan showed that the cyst had increased in size and it had developed mural nodules. She underwent a Robotic spleen preserving distal pancreatectomy. She had a prolonged recovery due to Grade B pancreatic fistula. Her final histopathology confirmed that the tumour was a dermoid cyst.

INTRODUCTION

Primary Pancreatic cystic neoplasms may be benign, premalignant, or malignant [1]. Serous cystic neoplasms, mucinous cystic neoplasms, and intraductal papillary mucinous neoplasms are the three most common types of primary pancreatic cystic neoplasms, representing about 90% of all primary pancreatic cystic neoplasms. Given that the biologic behavior of the different forms of primary pancreatic cystic neoplasm ranges widely, and because these patients are often asymptomatic, the detection of a primary pancreatic cystic neoplasm may represent a diagnostic dilemma for the practicing clinician [2]. Nonetheless malignant cystic tumors are rare whereas inflammatory cystic lesions are incredibly common [3].

A mature teratoma (dermoid cyst) of the pancreas is a rare congenital anomaly, which is believed to develop from epithelial inclusion, with persistence of embryonic tissue and pluripotent cells in the pancreatic parenchyma. Pancreatic teratomas probably originate from aberrant germ cells arrested in migration to the gonads early in embryonic life. The pancreatic location is one of the rare sites, and a review of the literature confirmed its rarity [4, 5, 6].

To our knowledge, the world literature reports 35 cases of pancreatic dermoid cysts with complete data [7].

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Abbreviations CT computed tomography; EUS FNA Endoscopic Ultrasound-Guided Fine Needle Aspiration; IPMS intraductal papillary mucinous neoplasm; MCN mucinous cystic neoplasm; PPCN primary pancreatic cystic neoplasm; SCNs serous cystic neoplasm
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CASE REPORT

A Seventy-five-year-old lady was found to have a complex cystic tumour of the tail of pancreas. A CT and MRI scan of the abdomen revealed a low attenuation lesion in the pancreatic tail measuring 2.9×3.1 cm. She had normal tumour markers and her clinical exam was normal other than COPD. An EUS FNA revealed a 20 mm complex solid mass in the tail of pancreas, which on aspiration looked like pus. The cyst fluid analysis could not be done, as the fluid was too viscous. The cytology showed low cellularity, occasional flat sheets of contaminant gastric epithelium admixed with cyst macrophages, debris, and occasional atypia. An interval CT over 3 months duration revealed that the pancreatic lesion had increased in size and additionally had enhancing mural nodules. For this reason she underwent a robotic spleen preserving distal pancreatectomy. Intra-operatively, the cyst looked benign, encapsulated, and free from surrounding structures. Surgical resection was done with minimal blood loss safeguarding the spleen and its vessels because of the unknown entity. She developed a grade B pancreatic leak that required EUS guided drainage and somatostatin analogue injections. Her final histopathology revealed that the cyst was lined by keratinizing squamous epithelium with underlying lymphoid stroma and numerous sebaceous glands. The presence of sebaceous glands confirmed the diagnosis of a benign dermoid cyst.

DISCUSSION

Pancreatic dermoid cysts are extremely rare tumors. Mature cystic teratomas of the pancreas were first described in 1918 by Kerr [8]. Vermeulen *et al*. reported a case in 1990, after having found 11 other cases between 1918 and 1977 [9].

Mature cystic teratomas are recognized by the presence of mature tissue elements from three germlines [4]. Those

reported in the pancreas are predominantly monodermal teratomas with only ectodermal derivatives and are referred to as dermoid cysts [10].

They are usually seen in younger patients (in their second or third decade of life). Clinical presentation of a pancreatic dermoid cyst is non-specific. Complaints at presentation include abdominal pain, back pain, nausea, vomiting, anorexia, weight loss, fatigue, fever and finally some cases can be diagnosed during a work-up for other diseases [11]. Jentschura *et al*. from Germany reported 16 cases in 1990. One of the patients in the series was found to have an asymptomatic calcified mass on routine chest x-ray. At the time of laparotomy, hair, sebaceous fluid, and a tooth were found within the cyst [12].

The difficulty in pre-operative diagnosis of pancreatic dermoid cysts is well emphasized in the literature even with the introduction of EUS [13]. The difficulty is due to the rarity of the lesion, but it may be suspected after imaging, like ultrasonography (US), CT, and MRI. The radiological appearances of these lesions are variable, depending on the proportions of the diverse tissues of which they are composed of [10, 13, 14].

An imaging classification system for these lesions has been proposed that is based on morphologic features of the lesion. Pancreatic cysts can be classified into four subtypes: (a) unilocular cysts, (b) microcystic lesions, (c) macrocystic lesions, and (d) cysts with a solid component [15].

Sonographically, the teratoma may be a predominantly cystic, solid, or complex masses, but as the echogenicity of fat overlaps that of other types of soft tissue, it is not possible to reliably identify fat on the basis of sonographic pattern alone [16].

Computed tomography is superior to sonography in the characterization and localization of teratomas. CT findings depend on the relative proportions of various tissues (solid, cystic and fat), which comprise the lesion. Calcifications are commonly seen in the solid component or in the wall. On CT scans, dermoid cysts are usually rounded, well circumscribed, extremely hypodense lesions with a Hounsfield unit measurement of -20 to -140, in keeping with their lipid content. A cystic tumour in the pancreas that contains fat and calcification may be suggestive of this diagnosis [16, 17, 18].

Fat-containing lesions have typically high signal intensity on T1-weighted images and on T2-weighted TSE images on MRI. Fat suppression or phase-shift GRE images have been recommended for differential diagnosis of lesions with high signal intensity on T1- weighted images [19].

EUS-guided fine needle aspiration (FNA) cyto-histolology has been shown to be a safe, reliable and cost-effective tool to detect pancreatic masses, but it does not allow an unquestionable diagnosis of nature and malignancy of the lesions [20].

A recent review by Antilon *et al*. about image-guided FNA biopsy of the pancreas revealed a relatively high overall sensitivity (64%–98%), specificity (80%–100%), and positive predictive value (98.4%–100%) [21]. In the literature, only three cases of preoperative diagnosis of dermoid cyst by FNA cytology were reported [22]. In 1991, Markovsky *et al*. described the findings of the first reported preoperatively diagnosed cystic teratoma by fine needle aspiration. Cytological findings included mature benign squamous cells, keratin debris and inflammatory cells and illustrated that such histological findings are inconsistent with other pancreatic disorders. The few previous descriptions of pancreatic dermoid cysts with FNA cytology described them as benign appearing with mature squamous cells, inflammatory cells and keratin debris [13]. It is important for pathologic evaluation of dermoid cysts to include complete sampling of the cyst wall in order to exclude the presence of immature foci (most commonly neuroepithelial type) as 7-10% of other retroperitoneal teratomas have been reported to be malignant [23, 24].

Fine-needle aspiration can be used to determine tumor pathology by using a variety of indices including cytology, flow cytometry, viscosity, or amylase, lipase, carcinoembryonic antigen, CA 15.3, CA 19–9, or CA 125 [25]. Lewandrowski *et al*. prospectively analyzed cyst fluid content of 26 patients whose pathologies were confirmed after surgical resection. They concluded that cyst fluid analysis can be helpful in making a diagnosis but that all cystic tumors should be resected whenever possible. Possible complications from percutaneous fine-needle aspiration include hemorrhage, infection, and spreading tumor cells [26].

On the other hand, some argue that cyst fluid analysis is not useful in the differential diagnosis from other pancreatic cystic neoplasms [14, 22].

Several factors influence the treatment of choice: pre-operative diagnostic accuracy, clinical presentation and type of symptoms, Location, type of resection and patient fitness. Mature cystic teratomas are strictly benign neoplasms and theoretically they do not require surgery. Nonetheless the application of the current guidelines for pancreatic cystic lesions [20, 27] does not obviate radical surgical resection for these lesions, because of inadequate preoperative diagnostic techniques. Cystic pancreatic tumors can be resected with an acceptable morbidity and mortality in a subset of patients as described in a series by Robert *et al.* [23].

In the case of symptoms or premalignant disease, attempts should be made to preserve the spleen, with either splenic vessel preservation (described by Kimura *et al*. [28]) or resection of the splenic vessels as described by Warshaw [29]. The advantages of preserving the spleen are the prevention of potentially lethal postsplenectomy sepsis and the use of vaccinations [30]. 125 patients who underwent distal pancreatectomy with or without splenectomy were involved in a retrospective study from the Memorial Sloan-Kettering Cancer Center. It revealed

that 49% of patients who underwent a splenectomy had post operative complications versus 39% of patients in whom the spleen was preserved. In addition, perioperative infectious complications and severe complications were more frequent in the splenectomy group (28 *vs*. 9% and 11 *vs*. 2, respectively) [31].

The robotic resection allows performing complex pancreatic resections with improved ergonomics, visualization, precision, and dexterity during spleen-preserving distal pancreatectomy. Furthermore, the dissection of the splenic vein and artery and the creation of the retropancreatic tunnel are more easily performed using the robot increasing the chance of splenic conservation [32]. Previous studies have shown that the use of Robot assisted surgery (RAS) is safe and efficient [33].

Various procedures such as simple excision, partial cystectomy,distalpancreatecomy,pancreaticoduodenectomy, internal drainage and external drainage have also been reported [34]. External drainage procedures have since been abandoned because of high rates of recurrence and fistula formation [35].

CONCLUSION

Cystic pancreatic lesions encompass a wide spectrum of non-neoplastic, benign, and malignant neoplasms and accurate characterization by imaging is made possible by adherence to imaging classification systems and management algorithms, which includes integration of laboratory and clinical data. However, their differential diagnosis can be quite challenging. It is important to recognize mucinous lesions because they often exhibit anadenoma-carcinoma sequence. We recommend strong index of suspicion when dealing with pancreatic cystic tumours, especially if complex, larger than 3 cm and if any sinister radiological findings. Resection should be the primary goal in managing cystic pancreatic tumors and there is evidence they can be resected with an acceptable morbidity and mortality [23, 36]. Favourable tumour location and careful patient selection could minimize operative mortality, morbidity and feasibility of minimally invasive surgery. Attempts to preserve the spleen should be made if the cysts are benign or premalignant as there are several advantages stated in literature [30]. RAS is a new and novel technique to facilitate this safely [32].

Conflict of Interest

The authors declare that they have no conflict of interest.

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