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Case Report

Acute post-traumatic presentation of a solid pseudo papillary tumor of pancreas: A case report

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Abstract

Objective: To report a case of pancreatic pseudo papillary solid cystic tumor, known as Frantz tumor, which was discovered after a blunt abdominal trauma and to carry out a review of literature.

Case report: The patient was a 14-years old girl who came to the emergency room after blunt abdominal trauma with an intense abdominal pain. She underwent emergent laparotomy because of the positive peritoneal signs and circulatory instability. The exploration revealed a large retroperitoneal mass. Therefore, an incisional biopsy was done and the patient was referred to our center.

CT-Scan was performed and revealed a large cystic mass (about 14×10cm) in left side of upper abdomen, which was located posterior to the stomach. Laparatomy revealed a very large pancreatic tumor with extension to spleen, stomach and transverse colon. A distal pancreatectomy, sleeve gastrectomy, segmental resection of large bowel and splenectomy was performed. The pathologic report was a pseudo papillary solid and cystic tumor of pancreas. Clinical evaluation has been satisfactory without recurrence after 12 months of follow up.

Conclusion: Frantz tumor has benign behavior and treatment of choice consists of surgical resection. Pseudo papillary tumor may spread outside the pancreas, particularly in peritoneal cavity. Met static spread may be promoted by trauma, including tumor biopsies which should never be performed. The outcome after surgical resection is excellent with 90% survival in the long term. Recurrence has been described in approximately 10% of the patients.

Key words: Trauma, solid pseudo papillary tumor of the pancreas

Introduction

Solid-pseudo papillary tumor of the pancreas is a rare condition in the literature and about 300 cases have been reported. This tumor was first described in 1959. It is also known under the term FRANTZ tumor, named after the author who first described it, which has also been referred to as cystic tumor; papillary epithelial neoplasia; solid and papillary epithelial neoplasia; or papillary epithelial tumor. The origin of the solid-pseudopapillary tumor has not yet been clarified. It is discussed to originate either from ductal epithelium (2, 4), acinar cells (19, 21), or endocrine cells (27-23). Another hypothesis is that this tumor arises from pluripotent embryonic cells of the pancreas (22, 24, 25) or from the ridge ovarian analogue related cells, which were attached to the pancreatic tissue during early emberyogenesis (22, 28). Solid-pseudopapillary tumor of the pancreas has a tendency to predominantly affect young woman aged between 25 and 35 years (3, 20, 24, 28, 29). Age is reported to range from 8 (25) to 70 year (2). However, a relationship with oral contraceptives has not been proven (20); this tumor rarely affects men and is characterized by a long asymptomatic course and nonspecific

symptoms. Therefore, it is not uncommon that solid-pseudo papillary tumor is detected only when it has been grown to a remarkable size of 8-10cm (3-20). Even a tumor of 20 cm in diameter has been reported (32). One feature of this tumor is its low malignant potential. Although the liver is found to be the mostly affected site by metastases, these are only rarely seen (20, 25, 32, 33). Furthermore, there are only few reports regarding invasive growth (2, 3). Survival time has been reported to reach up to 21 years (1).
Here we report a 14-ycar old woman with acute presentation of a solid pseudopapillary tumor of the pancreas who had suffered from a blunt abdominal trauma.

Case report

A previously healthy 14-years old girl suffered from a slight blunt trauma to the abdomen. The patient came to the emergency room of another center with intense atypical abdominal pain. An emergent laparatomy performed because of positive peritoneal signs and signs of circulatory instability. The exploration revealed a very large retroperitoneal mass. Therefore, a biopsy was done and the patient was referred to our hospital.
In history taking, she mentioned no weight loss or preexisting abdominal complain and swelling. Her vital signs were stable and chest and cardiovascular systems were normal. Abdominal examination revealed a mass occupying the epigastric and left hypochondriac region. Blood investigation revealed: Hemoglobin=14 g\dL (reference range: 11-15g/dL) and WBC=13×10^9/L (reference range: $3.5\text{-}10.0\times10^9$/L). Serum amylase and liver function tests were normal. Spiral abdominal and pelvic CT-Scan with oral and intravenous contrast showed a large cystic tumor (14×10cm) in the left side of upper abdomen. The tumor was located posterior to stomach and arising the body and tail of the pancreas. The cyst contained multiple enhancing solid components and showed mural enhancement. Linear areas of hyperdensities within the cyst suggesting hemorrhage were seen. Thus, cystadenoma of pancreas was the probable diagnosis; however, a pancreatic cyst could not be excluded. The patient went to the operation room with bowel preparation at recurrent laparatomy and we found a huge retroperitoneal mass with adherent to spleen, stomach and meso of the transverse colon. No metastases were found. We performed a distal pancreatectomy and partial gastrectomy, segmental resection of the large bowel and splenectomy. The weight of the specimen was 450 g.
Figure 3a, 3b: A macrograph of the resected operative specimen comprising distal of pancreas, a part of stomach, large bowel and spleen. Ten days later, the patient was discharged from our university hospital without any postoperative complications.
Histological assessment of sections showed residue of pancreatic tissue with an encapsulated cellular neoplasm composed of pseudopapillae covered by several layers of epithelial cells. The nuclei were ovoid and folded with indistinct nucleoli and few mitoses.
Figure 5a, 5b: Tumor cells radially around fibro vascular stalks forming rosette-1ike pattern. (H & E stain)
Figure 6: A micrograph showing cystic degeneration with solid and pseudopapillary formation and red blood cell in the cystic space.
The thick fibrovascular core of the papillae is composed of mucinous material with wide areas of tumoral necrosis. Solid pseudopapillary tumor (carcinoma with low malignant potential), $T2NOM_x$, stage IB, with tumor size measuring 20×l4×11cm was reported. Tumor had invaded the pancreatic capsule in some foci. All five excised lymph nodes were free of tumor. Vascular and perineural invasion was not identified. Gastric tissue and large bowel mucosa and surgical margins were free of tumor. The spleen showed hemorrhagic and infarction in one area.

Discussion

Solid-pseudopapillary tumor (SPPT) is the preferred term for a distinctive type of pancreatic tumor; also known as papillary and solid epithelial neoplasm, papillary-cystic neoplasm and cystic-solid papillary carcinoma.
Mostly, the cases are young women, and the most common clinical sign is a palpable abdominal mass. Grossly, it is usually large and on cross-section it often contains areas of hemorrhage and necrosis. Most cases are surrounded by a well developed capsule, but in some instances, the edges are those of a solid infiltrative neoplasm. Other cases are predominantly cystic and multicentricity is exceptionally rare. A few cases

have been found adjacent to but anatomically separate from pancreas (34). Mao in a cumulative review of the literature found that 90% of the patients were female with a mean age of 23.9 years (24). Another study in Taiwan between 1990 and 2003 reported seven patients with SPPT retrospectively reviewed, with a mean age of 31 years (57). Review of the literature revealed 24 cases of solid-pseudopapillary tumors of the pancreas reported in children (table1) with an average age of 10.8 years (range: 8-16 years) and a male: female ratio of 1:4.75 (5-18). There were 4 case (3 girls and a boy) 13-16 years of age presented acutely following blunt abdominal trauma in a fashion similar to that of our present case (5,11,14,15). An emergency whipple procedure was performed in 3 cases (11, 13, 15). Bombi et al. (54) reported other 2 cases of pseudopapillary tumors in older female patients 22 and 23 years of age, one presented with an acute abdomen and pneumoperitoneum.

Patients are often asymptomatic and the cyst discovered incidentally on physical examination or radiological studies(35). Patients may also occasionally present with a growing abdominal mass associated with vague abdominal discomfort or may rarely present with an acute abdomen due to tumor rupture and hemoperitoneum as happened in our patient. Jung et al. reported a series of 6 pediatric cases (4 girls and 2 boys) with a mean age of 11.2 years (range 8-13 years), 5 of the lesions were located in the pancreatic head necessitating pancreaticoduodenectomy and one was located in the tail which was treated by distal pancreatectomy (8). All were alive with no recurrence in an average follow-up of 5.5 years.

Wang et al. reported a solid-pseudo papillary rumor in 3 children (2 girls 11 and 14 years of age and a boy 10 years of age) and concluded that the tumor arises early in life, grows slowly and rarely metastasizes (6), Rebhandle et al. reported 4 girls 12-16 year of age presenting with abdominal pain and mass (diameter 7-15 cm), located in tail (n=2), the body and tail (n=1) and the head (n=l). Only one case developed two recurrences, and metastases were found in spite of surgical resection and adjuvant chemotherapy (9).

This tumor lends to be fairly benign in young females but appears more aggressive in older males whose mean age is about 10 years older than women (41, 50). Cilinically, patients with SPPT of the pancreas usually have vague abdominal symptoms with fullness or discomfort, pain, and a palpable abdominal mass and about 9% of the reported patients were asymptomatic (37).

Portsc reported a case of acute presentation of a SPPT in a young boy with a blunt trauma in whom a curative pancreaticodeodenectomy was performed (55). Another report was a 14 years old boy who underwent an emergent explorative laparatomy because of positive peritoneal signs and increased signs of circulatory instability. It revealed a large hematoma and the source of bleeding was a tumor arising from head of pancreas. Thus, a curative whipple procedure was

Table 1: Reported cases of solid pseudopaillary tumors of the pancreas in children

References	Year	Number of cases	Sex	Age(years)
Person et al. [5]	1996	1	Girl	16
Wange et al. [6]	1998	3	1 boy, 2 girls	10, 11, 14
Herskovits et al.[7]	1999	1	Boy	13
Jung et al. [8]	1999	6	2 boys, 4 girls	8-13
Rebhandl et al [9]	2001	4	Girls	12-16
Akiyama et al. [10]	2002	1	Girl	15
Cervantes-Monteil et al. [11]	2002	1	Girl	15
Sabatino et . al [12]	2003	1	Girl	15
Carrincaburu et. Al [13]	2003	1	Girl	13
Portc et. Al [14]	2003	1	Girl	9
Jiang et al [15]	2003	1	Girl	14
Saw et al [17]	2003	1	Girl	13
Andronikou et al. [16]	2003	1	Girl	15
Saw et al. [17]	2004	1	Girl	12
Bardales et al [18]	2004	1	Girl	13
Total		24	5 boys, 19 girls (Ratio 1:4.75)	Average: 10.8 years

accomplished. Physical examination is often normal a part from the presence of an upper abdominal mass (14).

In another case report, a young girl with diffuse peritoneal carcinomatosis due to pseudopapillary and solid tumor of the pancreas was reported. Fourteen months earlier, an abdominal trauma occurred.

Among 17 published cases of metastatic pseudopapillary and solid tumor of the pancreas (including 6 cases with peritoneal carcinomatosis), trauma was described in 11 cases (including 3 cases of peritoneal carcinomatosis). Pseudopapillary tumor may spread outside the pancreas, particularly in the peritoneal cavity. Metastatic spread may be promoted by trauma, including tumor biopsies which should never be performed (56). Usually, there is no evidence of pancreatic insufficiency. Abnormal liver function test, cholestasis, elevated pancreatic enzymes or an endocrine syndrome and tumor markers are all unremarkable.

Given the good prognosis of the disease, it is important to make the diagnosis preoperatively if possible, so that adequate resection will be undertake-no Therefore, imaging studies should be carefully assessed, with FNAC considered if necessary (37). Abdominal ultrasound and CT-Scan show a well encapsulated complex mass with both solid and cystic components and displacement of nearby structures. There may be calcifications at the periphery of the mass and intravenous contrast enhancement inside the mass suggestive of hemorrhage necrosis (36), Procacci C et al. reported the accuracy of CT scan in cystic pancreatic masses to be about 60% (41). According to cantisani et al. MRI is better than CT to distinguish certain tissue characteristics, such as hemorrhage, cystic degeneration, or the presence of a capsule, particularly as indicated by high signal intensity on Tl-weighted imaging and slightly progressive heterogeneous peripheral contrast Enhancement, seen after gadolinium administration on dynamic examination (42). Angiography usually Demonstrates a vascular or hypovascular pancreatic tumor and may help to delineate the mass from other involved and adjacent structures (37). Although, some radiological signs are suggestive of SPPT, radiological guided FNAC may be needed to obtain a preoperative diagnosis. In one study reviewing over 150 cases of SPPT, when preoperative FNAC was done, over 70% of lesions were definitely diagnosed as SPPT or had SPPT or low-grade epithelial neoplasm in the differential diagnosis (37). The origin of this tumor remains an enigma. Kosmahl (43) attempted to correlate the immunoprofile of tumors in 59 patients with a cellular origin for SPPT. They used different stains, including exocrine markers of acinar differentiation (trypsin, chemotropism), ductal differentiation (glycoprotein's), and neuroendocrine markers (synaptophysin and chromogranin). They found that the most consistent positive markers were vimentin, NSE, α-anritrypsin, α -amichymotrypsin and progesterone receptors, present in more than 90% of tumors. Cytokeratin was demonstrated in 70% and synaplophysin in 22%. However, their results failed to reveal a clear phenotypic relationship with any of the defined cell lines of the pancreas. Differentiation along exocrine cell lines has been postulated for SPPT on the basis of trypsin and chymotrypsin positivity. However, NSE and synaptophysin (43) positivity favors an endocrine origin. The female predominance along with the presence of progesterone receptors (43,45) in some reported cases suggests a neuroendocrine origin. In a study by Pezzi, SPPT had immunohistochemical and ultrastructural evidences of both an endocrine and intra-ductal differentiation, suggesting that this tumor may arise from a pluripotent stem cell (46). Although progesterone receptors have been found by some investigators, estrogen receptors have not been demonstrated (30, 45).

Another hypothesis by Kosmahl is that there is a close relationship between the pancreas and the genital ridges during embryogenesis, so that the tumor cells may be derived from the celomic epithelium and rete ovarii (43). These stem cells may become attached to pancreatic tissue during early embryogenesis (43,45). The differential diagnosis of SPPT of the pancreas includes any solid or cystic pancreatic disease entity, such as mutinous cystic tumor. micro cystic adenoma, islet cell tumor, cystadenocarcinoma, acinar cell carcinoma, inflammatory pseudo cyst, mucus secreting tumor, pancreatoblastoma, and a vascular tumor-like hemangioma. The first four are usually seen in older patients and have no particular gender preponderance (47).

Pancreatoblastoma is usually found in younger individuals of either sex. Radiologically, a linear sunburst pattern of calcification is the usual finding in micro cystic adenoma; a hyper vascular pattern on angiography is suggestive of islet cell tumor rather than SPPT (47).

Grossly, SPPT is a well-encapsulated, spherical mass, usually measuring around 8 to 10 cm. The cut surface shows large spongy areas of hemorrhage alternating with both solid and cystic degenerations. The histological appearance is very distinctive and is considered diagnostic. It is fundamentally a solid tumor with extensive degenerative changes forming solid cellular and hypervascular regions without glands. Areas of degeneration may then develop into Pseudopapillary structures. Nishihara (30) and colleagues showed that the presence of necrosis, vascular and perineural invasion, high nuclear grade, and prominent necrobiotic nests suggest a greater malignant potential and aggressive behavior. Another study reported a case of hepatic metastases in which there was DNA aneuploidy and elevated proliferative index (48).

One study demonstrated that even patients with local recurrence as well as liver and peritoneal metastases could still have long-term survival (49). Metastases reportedly occur at mean interval of 8.5 years (50). Surgery is the mainstay of treatment, which is usually curative for localized disease, There is evidence for prolonged survival after adequate surgical resection even with metastases, Even if the disease is extensive at the time of presentation, surgical debunking favors prolonged survival (51).

Intra-operative frozen section may be helpful to ascertain the adequacy of the resection margins. There have been only few reports of the use of radiotherapy (52) or chemotherapy (53), so it's difficult to judge the value of such measures.

In conclusion, SPPT of the pancreas is a rare indolent neoplasm with an unclear origin that typically occurs in young females. The diagnosis depends on histological confirmation, but its appearance on imaging is fairly characteristic, being a large well-encapsulated mass with calcification and areas of hemorrhagic degeneration. Surgical resection has generally been curative, but close follow up is recommended, particularly when the histologic appearance suggests a more aggressive tumor.

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