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Case report

Solid and papillary neoplasm of the pancreas (SPNP) – a rare pancreatic neoplasm detected by fine needle aspiration biopsy – a case report

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We present the case of a woman with a rare pancreatic tumor detected by fine needle aspiration biopsy guided by ultrasonography. The patient underwent left resection of the pancreas with splenectomy in 1989 and has been followed-up on an ambulatory basis since then. She remains in good general condition without any features of recurrent disease. SPNP detected in fine-needle aspiration biopsy points to the value of this method in the diagnosing of pancreatic tumors, including solid and papillary neoplasms of the pancreas.

Key words: solid and papillary neoplasm of the pancreas (SPNP), fine needle aspiration biopsy (FNAB)

Solid and papillary neoplasm of the pancreas is a rare pancreatic tumor that accounts for about 2.5% of primary tumors of the pancreas [1]. In the literature, 420 cases of SPNP in adults were reported until 2000 [2]. These tumors have been reported under various names, i.e. Frantz's tumor, Hamoudi tumor, solid and cystic tumor of pancreas, solid and papillary neoplasm, papillary-cystic epithelial neoplasm, papillary cystic and solid tumor of pancreas, and solid pseudopapillary neoplasm of the pancreas.

The first two cases were described in 1959 by Frantz [3].

SPNPs predominantly affect young women (mean age 25.6 years) with a 13 – 30-fold higher incidence compared with men [4- 6].

Most SPNPs do not present clinically. The commonest presentation is non-specific epigastric pain caused by compression of the neighboring organs by the tumor. This is reported by 45% of patients. A rare symptom is dyspepsia reported by 5% of patients. In 50% of patients SPNP presents as an epigastric mass detected in the course of physical examination or by imaging methods (USG, CT or MRI). Incidental diagnoses are rare and usually occur in patients after abdominal trauma who bleed to the abdominal cavity. Fine needle aspiration biopsy is a very helpful diagnostic tool. Its accuracy reaches 72% [7-9]. Detected tumors are usually over 10 cm in diameter [2, 3]. They are usually localized in the tail or the head of the pancreas (45% and 40%, respectively), less frequently in the pancreatic body [10]. Contrary to pancreatic cancer, SPNP rarely metastasizes (6%) and recurrences are also rare [11].

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Microscopically these tumors have a predominantly solid texture with microcysts and foci of necrosis. The most typical cytological feature of SPNP is the presence of papillary fronds composed of central fibrovascular stalks covered by layers of cuboidal or cyllindral tumor cells [9].

Immunohistochemical studies reveal that SPNP shows positive response for alfa-1 chymotrypsine, cytokeratine and CEA and negative response for the presence of neuroendocrine markers (neurofilament, PGP 9.5, chromogranine A, S100), protein p53, estrogen, and androgen receptors [8]. In the study of Tien et al. staining for progesterone receptors was positive in all 15 tumors analyzed by the authors (4 men and 11 women) but estrogen and androgen receptors were not detected. Nevertheless there were no gender-specific trends in the expression of sex-hormone receptor protein or in the clinicopathologic characteristics [5].

The treatment of choice is resection: left pancreatectomy with splenectomy (55%), Whipple (20%) or Traverso (10%) pancreatoduodenectomy or tumor resection (5-10%) [10].

We present a case of SPNP.

Case Report

In 1989 a 45-year-old woman (file no. 1511/89) reported pain in the left hypochondrium that had intensified during the last year. Her body weight did not change. Physical examination revealed a painless mobile mass about 15 cm in diameter in the left subcostal region. Ultrasound examination showed a polycyclic tumor, 12 cm in diameter, in the pancreatic tail. Percutaneous fine needle aspiration biopsy revealed solid and papillary epithelial neoplasm of the pancreas (SPNP). Abdominal CT confirmed the presence of the tumor, 12 cm in

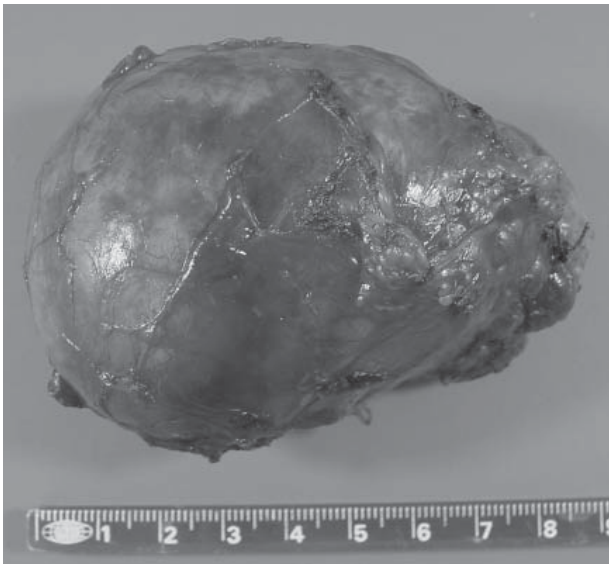

Figure 1. SPNP – the specimen after excision

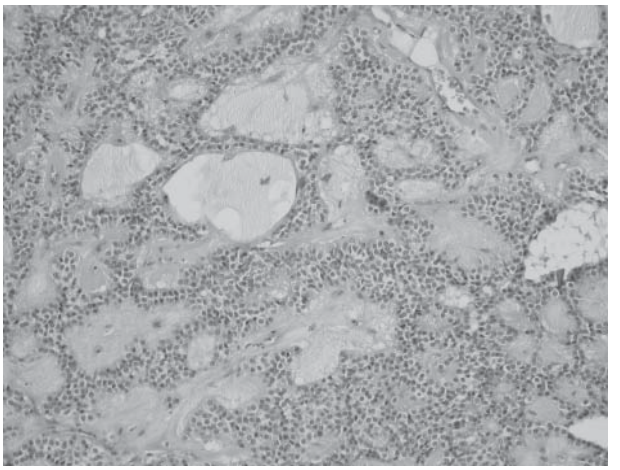

Figure 2. Histological features of the specimen. Characteristic pseudopapillary structures can be seen. Translucent hyaline globules are surrounded by tumor cells. Single neoplastic cells with enlarged nuclei and fine nucleoli are visible (hematoxilin-eosin, x 20)

diameter, in the pancreatic tail with foci of calcification and solid and cystic areas.

The results of laboratory tests were normal. The patient underwent resection of the pancreatic tail together with the tumor mass with splenectomy. The resected neoplasm (Figure 1) extended into the mesenterium of the transverse colon and to the omental sac.

The pancreatic stump was closed with single sutures, and drains were placed in the peritoneal cavity and omental sac. Postoperative recovery was uneventful. The patient was discharged on postoperative day 8 in good condition. Pathologically, the tumor was polycyclic measuring 14 x 9 x 6.5 cm. Microscopically, a solid and papillary neoplasm of the pancreas of low-grade malignancy was diagnosed (Figure 2). It showed positive response for α-1-chymotrypsin and negative response to glucagon, insulin, somatostatin and neuron-specific enolase. The sex hormone receptor status was not analysed in the study. The patient underwent radical resection (R0).

During follow-up no features of recurrence nor of metastases were found. After10 years the patient developed bilateral ovarian carcinoma and in August 1999 she underwent total hysterectomy with appendectomy and resection of the greater omentum. Histopathological examination confirmed bilateral ovarian carcinoma without metastases to omentum, a myomatous uterus and chronic appendicitis.

After surgery the patient received 6 doses of chemotherapy with Taxol and Cisplatin. She has been followed-up on an ambulatory basis and is in good general condition without subjective symptoms and with physical and additional tests within normal ranges.

Discussion

The presented case of solid and papillary neoplasm of the pancreas is one of two similar cases operated on in our Department of Surgery. The patient described in the study underwent successful radical operation without adjuvant therapy (radio- and chemo- therapy) and the long-term clinical follow-up confirms the benign course of the diseases as reported in medical literature [3, 9, 12]. The other case was that of a 37-year-old woman admitted for clinical jaundice and subsequently operated on. Intraoperatively it was found that the tumor infiltrated retroperitoneal vessels and thus it was not resected. Fine needle biopsy was performed twice during three years of follow-up. FNA material contained numerous cuboidal, polyhedral and cylindrical epithelial cells with eosinophilic cytoplasm. No mitotic figures were observed. Immunohistology revealed epithelial membrane antigen and α-1-chymotripsin in the tumor cells. Five years after the first operation the patient was operated on again due to acute cholecystitis – during the second laparotomy the pancreatic tumor was explored and frozen sections were performed, which allowed for the definite diagnosis of SPNP. The tumor was not resected and the patient was discharged home and remained in good condition [12].

Radical resection without adjuvant therapy is usually sufficient for SPNP. In all cases of SPNP the patients should be followed-up with clinical examinations, ultrasound, and CT scans.

In our case, the patient developed metachronous ovarian carcinoma 10 years after the onset of the primary disease. The second condition was considered incidental and not related to SPNP.

The diagnosis of SPNP by ultrasound-guided fine-needle aspiration biopsy points to the high diagnostic value of this method in detecting abdominal tumors.

High efficiency of fine needle aspiration biopsy has been confirmed in the cause of long-term observations by authors, who routinely used this method in the diagnosis of intraabdominal neoplasms [10, 13]. Histological verification of cystic and solid tumors is indicated to facilitate decisions concerning the treatment methods. Fine needle aspiration biopsy is safe, easily repeatable

with a low side-effects rate and a low mortality rate ranging from 0.01 – 0.17% mostly related to bleeding [13]. The only contraindication to fine needle aspiration biopsy in the opinion of the authors is the lack of patient cooperation and serious hemostatic disorders.

The other promising diagnostic modality to be used in case of pancreatic tumors is endoscopic ultrasound-guided fine needle aspiration biopsy (EUS-FNA). It is an excellent method of cytological diagnosis of samples from the pancreas, with a diagnostic accuracy of more than 90% for pancreatic adenocarcinoma. EUS-FNA is currently used for the preoperative diagnosis of pancreatic cysts and for small neoplasms. EUS-guided aspirates are usually contaminated by epithelial cells from the stomach, which may complicate the diagnosis. Its value in diagnosing intraductal papillary mucinous neoplasm (IPMN) of the pancreas was emphasized by Salla Ch.et al. They concluded that close cooperation between an experienced endoscopist and cytopathologist may result in an accurate diagnosis of IPMN, based on EUS-guided FNA cytology [14]. EUS-guided FNA cytology emerges as an accurate method in the preoperative diagnosis of pancreatic lesions and may be helpful in proper identification of solid and papillary neoplasm of the pancreas. The assessment of this modality requires further studies.

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