CASE REPORT

Mucoepidermoid carcinoma of the pancreas: a case report and a review of literature

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Mucoepidermoid carcinoma of the pancreas is rare. Here, a 63-year-old woman with left upper abdominal pain and abdominal distension is presented. Her mucoepidermoid carcinoma was located at the left upper abdomen, arising from the pancreatic body and tail without invasion of pancreatic capsule. On pathologic examination, the tumor consisted of three types of cells, the majority being poorly differentiated adenocarcinoma cells with mucin products in their cytoplasm, and some moderately differentiated adenocarcinoma with a tendency to form ducts. In addition, there were epidermoid cells and intermediate undifferentiated cells. She survived for 12 months after surgery.

Keywords: Mucoepidermoid carcinoma, pancreas, splenic artery aneurysm, squamous carcinoma, adenosquamous carcinoma

INTRODUCTION

Mucoepidermoid carcinoma was first described as a separate entity in 1945 by Stewart *et al*.[1] and is most commonly found in the salivary glands,[2-4] bronchi,[5-9] and liver.[10,11] However, mucoepidermoid carcinoma of the pancreas is rare. The first documented case was reported in 1959 by Franz.[12] To our knowledge, a total of seven cases[12,13] have been reported, and we report herein the eighth case and review the literature on the previous cases.

CASE REPORT

A 63-year-old woman was admitted to our hospital, following a 5-year history of left upper abdominal pain and abdominal distension (The Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China, 2011). Computed tomography (CT) scan performed by a local physician 1 week before admission revealed suspected pancreatic cancer. She was then referred to our hospital for further examination and treatment. At the time of admission, she complained of loss of appetite and nausea, but no jaundice was found. Her abdomen was slightly distended and tympanitic. No remarkable mass was found in abdomen by physical examination. The serum levels of carbohydrate antigen 19-9 (CA19-9) was elevated at 500.1U/ml (with normal range less than 37 U/ml). The serum level of carcinoembryonic antigen (CEA), squamous cell carcinoma antigen, alpha-fetoproteins, and neuron-specific enolase were not elevated. Computed tomography scan revealed a solid mass, approximately 4.5 cm in diameter, located at the left upper abdomen. The tumor was irregular in shape, and appeared to arise from the distal pancreas [Figure 1A]. Meanwhile, a splenic artery aneurysm existed [Figure 1B].

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Operative findings

A laparotomy was performed and we found that the tumor arised from the pancreatic body and tail without breaking pancreatic capsule. Meanwhile, a splenic artery aneurysm was found near celiac trunk. The distal pancreas, spleen, and its aneurysm were resected [Figures 2A and 2B]. Intraoperative frozen section of the specimen showed the proximal pancreatic margin was negative.

Pathologic examination

On pathologic examination, the size and the cut surface showed a milky-white solid and homogeneous tumor with a hard elastic consistency. The tumor consisted of three types of cells, the majority being poorly differentiated adenocarcinoma cells with mucin products in their cytoplasm, and some moderately differentiated adenocarcinoma with a tendency to form ducts. In addition, there were epidermoid cells and intermediate undifferentiated cells. These epidermoid cells did not

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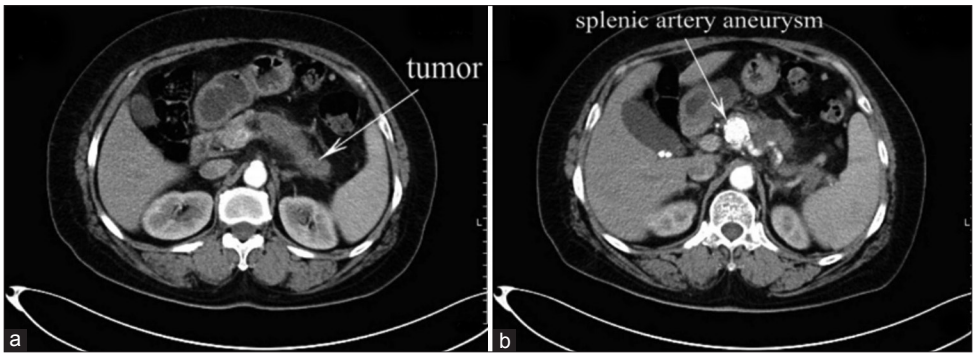

Figure 1: (a) Transabdominal CT revealed a solid mass, approximately 4.5 cm in diameter. The tumor was irregular in shape, and appeared to arise from the distal pancreas. (b) A splenic artery aneurysm existed near celiac trunk

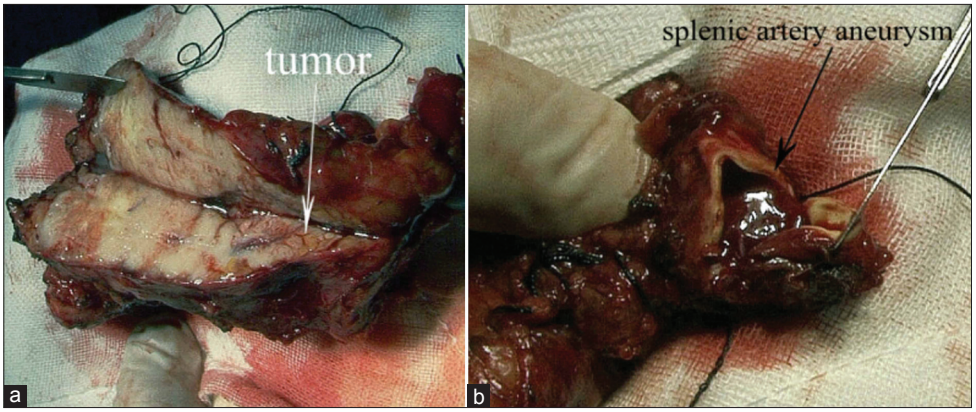

Figure 2: (a) The specimen removed from the pancreas, the tumor arised from the distal pancreas without breaking pancreatic capsule. (b) The specimen of the splenic artery aneurysm was near celiac trunk

have definite intercellular bridges, keratohyaline, or keratin pearls. No differentiated squamous carcinoma cells were detected in any part of the tumor [Figure 3].

Immunohistochemical findings

The cells of the tumor showed strong staining with antibodies to CK7, CK5/6, and CEA, showed negative staining for CK20, the proliferation index by Ki-67 stain was above 30%. The final pathology diagnosis was mucoepidermoid carcinoma. The postoperative course was uneventful and the patient was discharged 2 weeks after the surgery. Gemcitabine was used once but the patient had severe gastrointestinal side-effects, then she underwent oral Xeloda for five cycles. However, CA19-9 and CEA were elevated 5 months after operation. She survived for 12 months after surgery.

DISCUSSION

The first documented case of mucoepidermoid carcinoma of pancreas was reported in 1959 by Franz. The pathogenesis of this tumor is yet unknown. Ohtsuki *et al.* suggested four hypotheses to account for the pathogenesis of the squamous component in pancreatic cancer, namely: (1) from pluripotent undifferentiated cells into mucin-producing cells and/or squamous cells; (2) from ectopic squamous cells; (3) from metaplastic squamous cells; and (4) from the squamous metaplasia of underlying adenocarcinoma.[14] In our report, the mucoepidermoid cells stained positively for CK7, CEA, and CK5/6, but negatively for CK 20. Based on the CK immunoprofile, considering the present case, the appropriate hypothesis is the fourth. The staining for Ki-67 was above 30% and demonstrated possibility of a poor prognosis. Thus, a close follow-up is needed, to allow early treatment in case of recurrence.

CT or US (Ultrasound) imaging is hard to distinguish mucoepidermoid carcinoma from the other types of pancreatic tumors before operation. For our case, though the mass can be identified on the body and tail of pancreas, we cannot judge the specific from the preoperative imaging. It behaves unpredictably, with the majority possessing some metastatic potential.

Only seven cases of mucoepidermoid carcinoma of the pancreas have been reported, except that of our patient [Table 1]. The ages of the patients ranged from 48 to 69 years, with the median age of 60.5 years, and five were male and three female. The tumors were located at different sites of the pancreas (two at the head, three at the body, two at

Table 1: Cases of mucoepidermoid carcinoma of the pancreas

Case	Sex	Age (year)	Location	Diameter (cm)	Progression	Operation	Prognosis
Li JT *et al.* (2012) Current case	F	63	Body and Tail	4.5	Local pancreatic tumor	Pancreatic body and tail resection	12 months, dead
Li XD *et al.* (2002)[13]	M	65	Body	10	Local pancreatic tumor	Not available	Not available
Onoda N *et al.* (1995)[12]	M	64	Tail	8	Invasion to spleen, left kidney and adrenal, colon, multiple liver metastases, peritoneal dissemination	Pancreatic tail resection	11 months, alive
Kimura *et al.* (1993)[12]	M	57	Body	8	Invasion of stomach, colon, multiple liver and lung metastasis	Intraoperative radiation therapy	2 months, dead
Hayashi *et al.* (1992)[12]	M	58	Head	3.5	Lymph node metastasis	Pancreatoduodenectomy	6 months, alive
Kishimoto *et al.* (1991)[12]	F	48	Body	3.5	Solitary liver metastasis	Total pancreatectomy	11 months, dead
Ohtsuki *et al.* (1987)[14]	M	58	Tail	10	Multiple liver metastases	(-)	2 months, dead
Ohshio *et al.* (1987)[12]	F	69	Head	T2	Invasion of mesocolon, superior mesenteric vein	Pancreatoduodenectomy	3 months, dead

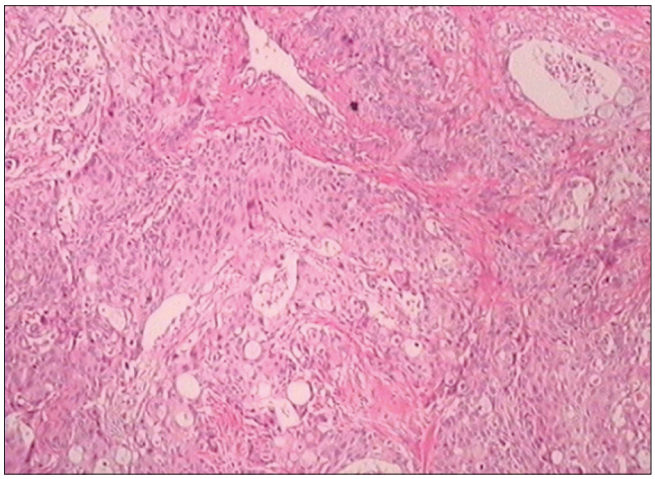

Figure 3: (H and E, ×100) The tumor consisted of three types of cells, the majority being poorly differentiated adenocarcinoma cells with mucin products in their cytoplasm, and some moderately differentiated adenocarcinoma with a tendency to form ducts. In addition, there were epidermoid cells and intermediate undifferentiated cells

the tail, and one at the body and tail) and were all larger than 3.5 cm in diameter at the time of diagnosis. All cases were advanced and every tumor grew invasively except our case, with six of the eight cases showing metastatic lesions in the liver and/or lymph nodes. The prognosis was extremely poor in all cases. Only one case was survival more than 11 months in previous cases and our patient merely survived for 12 months. Treatment consisting of surgery and radiation proved not to be very effective.[12] Based on the experience of this case, surgery seems to be only modality which brings the hope of cure if the mass is resectable.

Mucoepidermoid carcinoma is thought to be a subtype of adenosquamous carcinoma, histologically characterized by the presence of two components: an adenocarcinoma and a squamous carcinoma,[15,16] but Stewart *et al.* declared that mucoepidermoid carcinoma has a different histological character than adenosquamous carcinoma. Mucoepidermoid carcinoma is composed of three types of cells, including mucin-producing cells, epidermoid cells, and cells intermediate between basal cells and epidermoid cells, mixed in various proportions.[1]

As far as mucoepidermoid carcinoma of pancreas be concerned, ordinary tubular and/or papillary adenocarcinomatous structure would not be detected in the pancreatic lesion, and cancer cells can produce mucin. Ultrastructural examination shows that some of the cancer cells possess intracytoplasmic lumina surrounded by

bundles of tonofilaments, squamous, and undifferentiated cells with transitional forms.[14] Our case should be designated as mucoepidermoid carcinoma rather than adenosquamous carcinoma which are composed of mucin-producing cells, epidermoid cells, and cells intermediate between basal cells and epidermoid cells.

CONCLUSIONS

In summary, mucoepidermoid carcinoma of pancreas cannot be differentiated from pancreatic adenocarcinoma by the preoperative examination, and surgery is still the only effective treatment if the mass was resectable. Due to limited cases of this disease reported, the effective diagnostic and therapeutic approach cannot be figured out. Therefore, more experience should be anticipated.

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