CASE REPORT

Retropancreatic Ovarian Tumor

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Received: 18 March 2015 / Accepted: 27 October 2015 / Published online: 12 November 2015
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Abstract Retroperitoneal mucinous cystadenomas are rare lesions (less than 50 reported) characterized by presence of ovary like stroma of unknown origin. However, germinal component of ovary has never been found in them. The pancreas occasionally gives rise to mucinous cystadenomas, but they are always intrapancreatic. We report a unique case of a rare retroperitoneal mucinous cystadenomas with presence of ovarian follicles in a 45-year-old lady who presented with an abdominal mass. This was successfully excised. Though retroperitoneal mucinous cystadenomas are rare, presence of ovarian follicle (germ cell) in them has never been reported before.

Keywords Retropancreatic tumors · Retroperitoneal tumors · Mucinous cystadenomas · Mucinous cystadenocarcinoma

Case Report

A 45-year-old lady presented with gradual upper abdominal distension with associated sensation of fullness after meals. There was no significant comorbidity and no past history of surgery. Patient had a normal menstrual history and had two children born from a normal delivery. Examination revealed mild pallor; no icterus, fever, lymphadenopathy, or pedal edema. Abdominal examination revealed a large mass (almost a

foot across), in the midline, in the upper abdomen; its upper borders were not palpable and it moved quite well with respiration. There was no other organomegaly.

A CT scan (Fig. 1) revealed a septate, solid cystic mass in the region of the tail of the pancreas/splenic hilum. Uterus and ovaries were normal. Routine preoperative investigations revealed nothing significant except a hemoglobin level of 9 g/dl.

On exploration, a 4-kg, 30-cm, ovoid tumor (Fig. 2a, b) was found in the retroperitoneum, posterior to pancreas. It had a clear plane of dissection around it, though the pancreatic body and tail was splayed on its anteromedial surface. There were some distended veins on the surface without a definite pedicle. There was no evidence of local invasion, lymphadenopathy, or liver metastasis. Uterus and ovaries were normal. Though the pancreas could be separated quite cleanly from the lesion, we left a drain in the tumor bed, which we removed on day 4 post-op.

The histopathology report was “papillary mucinous cystadenoma”; the tissue also revealed ovarian stroma and corpora albicans (ovarian follicles) which was never reported before in these types of tumors (Fig. 2c).

Discussion

Retroperitoneal mucinous cystadenomas are rare lesions, with less than 50 reported till date [1]. They usually present with mass effect. Sometimes, pain and intermittent intestinal occlusion also occurs [2].

Preoperative diagnosis is difficult. Radiologically, retroperitoneal mucinous cystadenoma presents as a unilocular or multilocular cyst pressing the surrounding organs. The differential diagnosis of retroperitoneal mucinous cystadenoma is

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Fig. 1 CT scan showing the tumor

made with lymphangioma, cystic teratoma, lymphocoele, urinoma, and cystic mesothelioma [3]. CT revealed a mass in the region of the tail of the pancreas/splenic hilum, $30 \times 15.5 \times 11$ cm, showing intramural calcification typical of cystadenoma. The latter helps in the differential diagnosis between cystic teratoma and cystadenoma. Calcifications found within the mass support the diagnosis of teratoma while mural calcifications confirm the diagnosis of cystadenoma.

There is no specific tumor marker (e.g., CA-125, CA-19.9, and CEA) [2]. On gross examination, tumor may be uni- or multicystic containing gelatinous material. Microscopically, what characterizes them is the presence of ovary like stroma, the origin of which is uncertain. Hypotheses include development from an “ectopic ovary” or persistence of developmental müllerian elements or a monodermic teratoma with mucinous

epithelium proliferation [4–6]. Currently, theory of metaplasia in overlying coelomic epithelium is accepted. During the embryogenesis, coelomic epithelium converts to peritoneal mesothelium and ovarian germinal epithelium. Peritoneal mesothelium seems to keep the same differentiation properties as ovarian germinal epithelium. The similarity of immunohistochemical and ultrastructural profiles with ovarian mucinous tumors supports this hypothesis [2, 6]. The immunohistochemistry analysis may show a positive match to CK7 and CK20 antibodies. This is the same profile encountered in ovarian mucinous tumors. However, the cases reported so far have not shown any germ cell (ovarian follicle) derivatives [7, 8]. The fact that this tumor is also occasionally found in men and ovarian follicle (germ cell derivative) has not been detected previously has made the “ectopic ovary” theory weak [7].

In our patient, we found corpora albicans representing atrophic follicles, a germ cell derivative. This makes our case unique. It supports the theory that tumor’s growth is due to an ectopic or aberrant ovarian tissue.

The pancreas also gives rise to mucinous cystadenomas, which are in fact the commonest of its cystic neoplasms. These lesions are characterized by presence of “ovarian stroma” to the extent that it has been proposed that these tumors be recognized by this very feature [8]. However, such tumors are intrapancreatic and also they never contain any ovarian follicle.

Surgical removal by open or laparoscopic approach is the treatment of choice. Surgery not only alleviates mass effect but also prevents future risk of infection or malignant change. A meticulous sampling of the resected tumor is necessary to search for rare occurrence of borderline malignancy or a carcinomatous focus [2]. In women with focus of carcinoma, some authors suggest ovariectomy or hysterectomy [7].

Fig. 2 a–c Intraoperative picture, specimen picture, and histopathology picture (H & E stain; original magnification $\times 40$, showing papillary formation by columnar epithelial cells secreting mucin and corpora albicans)

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no competing interests.

Research Involving Human Participants and/or Animals Not applicable.

Informed Consent Informed consent was obtained from all individual participants included in the study.

Ethical Approval For this type of study, formal consent is not required. This article does not contain any studies with animals performed by any of the authors.

Funding None.

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