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Case Report

Serous cystadenocarcinoma of the spleen

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ABSTRACT

The commonly seen primary malignant neoplasms of the spleen are angiosarcoma and lymphoma. We present a case of serous cystadenocarcinoma of the spleen. It was presumed to be originated from dropped nonmalignant ovarian tissue, which was accidentally implanted to the splenic surface during hysterectomy and bilateral salpingooophorectomies for torsion of right fallopian tube 9 and half years ago and transformed into serous cystadenocarcinoma later. Computed tomography demonstrated a multilocular predominantly cystic tumor with internal soft tissue components in the spleen.

Introduction

Primary neoplasms of the spleen include lymphoma, angiosarcoma, hemangioma, lymphangioma, littoral cell angioma, hamartoma, and inflammatory pseudotumor [1]. Other unusual tumors include lipoma, angiomyolipoma, fibroma, fibrosarcoma, leiomyosarcoma, malignant fibrous histiocytoma, and mucinous cystadenocarcinoma [1,2]. The authors report a serous cystadenocarcinoma solitarily present in the spleen and discuss its probable origin.

Case report

A 65 year-old female consulted the internal medical department with a chief complaint of epigastralgia for several months. She received hysterectomy and bilateral salpingooophorectomies 9 and half years ago due to torsion of right fallopian tube. The gross specimens showed torsion of right fallopian tube with swelling and gangrenous change and congestion of right ovary. Bilateral ovaries and left fallopian tube were of normal outlooking and size. The microscopic examination showed hemorrhagic necrosis of left fallopian tube. It did not reveal any malignancy in the uterus, fallopian tubes, and ovaries. This time, the epigastric pain progressively increased in severity and became constant. An upright abdomen film showed splenomegaly. A subsequent computed tomography examination demonstrated a multilocular cystic tumor, about 16.0 cm in greatest dimension, with irregular internal soft tissue components in the spleen (Figs. 1A and B). The patient was admitted to the surgical ward to receive further evaluation.

Competing Interests: The authors have declared that no competing interests exist.

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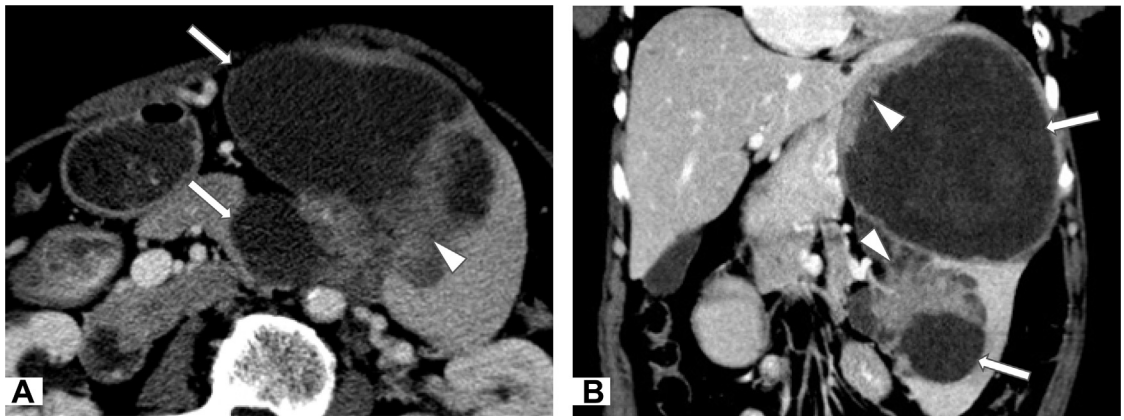

Fig. 1 – (A) Postcontrast axial image and (B) postcontrast reformatted coronal image. A multilocular predominantly cystic tumor with relatively smooth tumor wall (arrows) and internal soft tissue components (arrowheads) was seen.

A percutaneous biopsy revealed clusters of moderately differentiated neoplastic cells bearing hyperchromatic nuclei, prominent nucleoli, and vacuolated cytoplasm and arranged in focal papillary pattern. The mucicarmine stain was positive. Immunohistochemically, the tumor cells were positive for cytokeratin (CK) 7, but not for CK20, CDX2, and thyroid transcription factor (TTF)-1. The results suggested a metastatic adenocarcinoma. The serum levels of cancer antigen (CA) 15-3 and CA 125 were 69.3 U/mL (normal <31.3U/mL) and 2352.8 U/mL (normal <35U/mL), respectively. Those of carcinoembryonic antigen (CEA) and CA 19-9 were within normal range. The following surveys in chest, breast, and upper and lower gastrointestinal tracts did not detect any possible primary malignancy. A surgical intervention was performed to remove the spleen. During operation, the spleen was easily isolated and resected without any adhesion to the adjacent organs. The gross specimen (Fig. 2) revealed a multilocular cystic tumor with internal soft tissue components, necrotic tissue, and hemorrhage. There was no detectable mucin content within the tumor. The microscopic examination (Figs. 3A and B) demonstrated clusters of moderately differentiated neoplastic cells bearing hyperchromatic and pleomorphic nuclei, prominent nucleoli, and eosinophilic cytoplasm, which were arranged in papillary pattern. There were multiple psammoma bodies disseminated in the tumor. No mucin-secreted epithelium was found. Immunohistochemically, these tumor cells were positive for CK-7 (Fig. 3C) and Ber-EP4 but negative for CK-20 (Fig. 3D), CDX2, and TTF-1. These results were consistent with serous cystadenocarcinoma [3]. A positron emission tomography examination 1 month later did not detect any other possible malignancy. Based on these findings, a diagnosis of serous cystadenocarcinoma of the spleen was made. The patient was regularly followed up in outpatient department. One and a half year later, a palpable left neck lymph node was noted. The biopsy showed malignant cells with same immunochemical stains, including positive CK-7, negative TTF-1, CK-20, and CD20 stains, as that of splenic serous cystadenocarcinoma. The result was judged as lymph node metastasis. The patient received local adjuvant radiotherapy of the neck and regular follow-up.

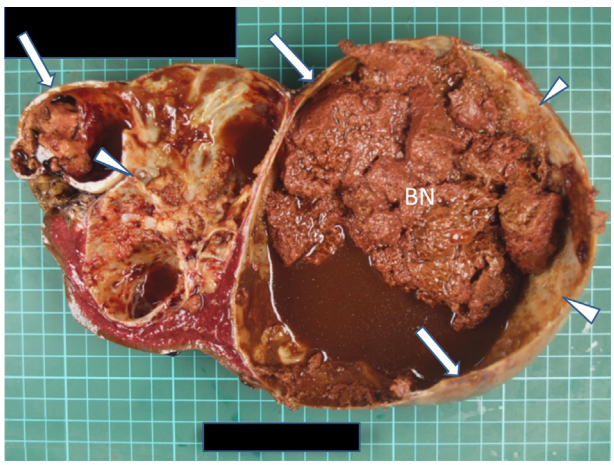

Fig. 2 – The gross specimen showed the cystic tumor wall (arrows), internal soft tissue components (arrowheads), and a mixture of blood and necrotic tissue (BN).

Discussion

Primary neoplasms of the spleen include lymphoma from lymphoid tissue and hemangioma, lymphangiom, hamartoma, littoral cell angioma , hemangioendothelioma, hemangiopericytoma, and angiosarcoma from vascular element [1]. Primary cystadenocarcinoma of the spleen is extremely rare. All the reported cases were mucinous type [2,4–9]. The hypothesized origins of these tumors include (1) ectopic pancreatic tissue; (2) ectopic intestinal tissue; and (3) invaginated mesothelium of the splenic capsule [2,6]. In contrast to the mucinous type, the current case might represent a different entity. The patient received hysterectomy and bilateral salpingooophorectomies 9 and half years ago due to torsion of right fallopian tube. Neither gross