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Mature ovarian cystic teratoma with disseminated nodular lesions in the pleural and peritoneal cavities: A case report

Takamichi Minato MD[a,b], *Masafumi Toyoshima MD, PhD*[a,*], *Noriaki Imai MD, PhD*[b,1], *Akiko Kasai MD, PhD*[b], *Nobuo Yaegashi MD, PhD*[a]

[a] *Department of Obstetrics and Gynecology, Tohoku University Graduate School of Medicine, 1-1 Seiryo-machi, Aoba-ku, Sendai, Miyagi 980-8574, Japan*

[b] *Department of Obstetrics and Gynecology, Hachinohe City Hospital, Aomori, Japan*

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ABSTRACT

Mature ovarian cystic teratoma (MOCT) is the most common benign neoplasm of the ovary and has a wide spectrum of radiological presentations. Our aim was to present the radiological characteristics and pathologic findings of a patient with an atypical manifestation of this common disease. A 52-year-old Japanese woman was admitted to our hospital with a large cystic mass in the pelvis and an elevated squamous cell carcinoma antigen level. Computed tomography revealed disseminated cystic lesions in the intraperitoneal and intrathoracic spaces. The lesions contained fat and featured calcifications. Laparotomy revealed many white, spherical nodules in the peritoneal cavity. The results of pathologic examination led to a presumed diagnosis of a foreign body reaction to the contents of an MOCT that leaked into the peritoneal cavity. The patient has been followed up for 13 months and remains free of symptoms without additional treatment. We describe a rare presentation of MOCT, in which we initially strongly suspected an advanced malignancy based on the results of imaging. To make an accurate diagnosis, it is necessary to understand the rare findings associated with MOCT, as well as the common signs on different imaging modalities.

Introduction

Mature ovarian cystic teratoma (MOCT), also known as ovarian dermoid cyst, is the most common benign neoplasm of the ovary [1]. Teratomas are a type of germ cell tumor that contain well-differentiated embryologic tissues from more than 1 germ cell layer (ectoderm, mesoderm, and endoderm) [2]. Teratomas are slow-growing, encapsulated tumors; spontaneous rupture is uncommon [3]. Because of the heterogeneity of MOCT,

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[1]Present address: Department of Obstetrics and Gynecology, Sendai City Hospital, Miyagi, Japan.

* *Corresponding author:*
E-mail address: m-toyo@med.tohoku.ac.jp (M. Toyoshima).

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patients demonstrate a wide spectrum of radiological presentations [4]. The differential diagnosis may include a malignant ovarian cancer of advanced stage when the mass has ruptured into the intraperitoneal space [5]. The aim of this case report was to present the radiological findings of MOCT with disseminated nodular lesions in the peritoneal and pleural cavities, with a brief review of our patient's pathologic findings.

Case report

A 52-year-old Japanese woman was admitted to our hospital for evaluation of an ovarian cyst. She had been told that multiple cysts were present within her abdomen. She denied any digestive or gynecologic symptoms. She had a history of surgery for appendicitis at 7 years of age and had taken medication for hypertension from the age of 48 years. She smoked 13 cigarettes per day for the past 32 years and drank alcohol every day. She had been pregnant 9 times with 8 vaginal deliveries and 1 spontaneous abortion. Physical examination and ultrasonography (USG) revealed a large cystic mass with diffuse internal echo in the pelvis (Fig. 1A) and a 5-cm fibroid on the posterior wall of the uterus. There were several calcified cysts with a strong acoustic shadow in the abdomen (Fig. 1B). Her laboratory results were within normal limits, including cancer antigen 125 and cancer antigen 19-9 levels. The only test that was abnormal was an elevated squamous cell carcinoma (SCC) antigen level (6.4 ng/mL).

Contrast-enhanced computed tomography (CT) showed an encapsulated cystic mass containing a focal, solid, enhancing nodule component located in the pelvis (Fig. 1C). Multiple, variable-sized disseminated cystic lesions featured ringlike calcifications with distinct borders were found in the abdominal cavity (Fig. 1D), the liver, and the omentum (Fig. 1E). There were many small, nodular thickenings under the pleura indicating old pleuritis (Fig. 1F). Magnetic resonance imaging (MRI) showed a multicystic mass approximately 16 cm in diameter in the pelvic cavity, and the periphery of the mass was hypointense on T2-weighted imaging (Fig. 1G). The tumor content showed a marked signal loss on fat-suppressed T1-weighted MRI, indicating a fatty lesion at the center of the cyst (Fig. 1H). The mass lesion showed no contrast enhancement on either CT or MRI. Although there was no clear finding suggesting malignancy in diagnostic imaging, we suspected malignant transformation of a mature ovarian teratoma because of a large number of seeding-like lesions in the peritoneal cavity and the increased SCC antigen level.

Laparotomy revealed that the main tumor was derived from the right ovary. There were many white nodules in the peritoneal cavity (Fig. 2A). The results of intraoperative pathology indicated that the right ovarian tumor was a mature cystic teratoma and the intraperitoneal nodes were degenerated tissue without malignant findings. We performed a total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, partial liver resection, partial diaphragmatic excision, and removal of the abdominal nodules (Fig. 2B).

Pathologic examination revealed that the ovarian lesion was a cyst filled with a gelatinous viscous material and hair, typical of a mature cystic teratoma (Fig. 2C). Skin, bone, cartilage, bronchial epithelium, mixed glandular tissue, and mature brain tissue were observed, but there were no malignant cells or immature tissue types in the surgical specimen. Nodules in the omentum consisted of coated structures composed of thick, glass-like collagen tissue (Fig. 2 D, E). It was presumed that the nodular lesions disseminated in the peritoneal cavity were generated by a foreign body reaction to leakage of the MOCT. The patient was followed up for 13 months and remained free from symptoms without additional treatment.

Discussion

Patients with MOCT demonstrate echo-complex tumors on USG, with cystic and solid components and echogenic spots with acoustic shadows [4]. The most common sonographic finding is the Rokitansky nodule, which is seen as a densely echogenic nodule protruding into the cyst [4]. If contrast enhancement is observed at the site of a Rokitansky protuberance, malignant transformation is likely [1]. About 1%-2% of mature teratomas reportedly undergo malignant transformation [6], with risk factors including patient age of >45 years and an SCC antigen level of >2 ng/mL [7]: both were observed in our patient. This is why we suspected the malignant transformation of MOCT from chronic granulomatous peritonitis before laparotomy, although it was no malignant finding such as contrast effect in diagnostic imaging.

The most characteristic radiological findings of MOCT are a complex mass containing a well-circumscribed fluid component of variable volume, the presence of adipose tissue or sebum appearing as a fat-fluid level, and calcifications in either a congealed or a linear-strand pattern [8]. These findings are better demonstrated by CT than by USG. In fact, CT has excellent sensitivity for detecting MOCT [4] [9] because CT can distinguish the component of lesions by providing information regarding density measurements. The presence of fat, calcification, hair, and Rokitansky protuberance demonstrated by CT imaging is diagnostic in most of MOCT cases. Fat density numbers in MOCT are reported as ranging from −144 to −20 HU [10]. On the other hand, MRI is preferred to be used for difficult cases or when malignancy cannot be denied. MRI provides information necessary for the provisional diagnosis of teratoma, as it is very sensitive for the fatty and calcific elements commonly represented in teratomas. Almost all lipid-containing masses within the adnexa are ovarian teratomas. A sebaceous component in an MOCT can be identified using very high signal intensity on T1-weighted images and signal dropping in fat-suppressed T1-weighted images [2]. Rupture of MOCT occurs in 1%-4% of patients [11]; the clinical presentation of rupture may take either an acute or a chronic course. An acute presentation may be caused by severe chemical peritonitis associated with acute abdominal crisis and shock. Acute peritonitis is caused by sudden rupture of the tumor contents during torsion, which results from infection, rapid growth of the cyst, direct trauma, or prolonged pressure during pregnancy [3,11]. In contrast, chronic rupture is caused by slow and repetitive leakage from a small

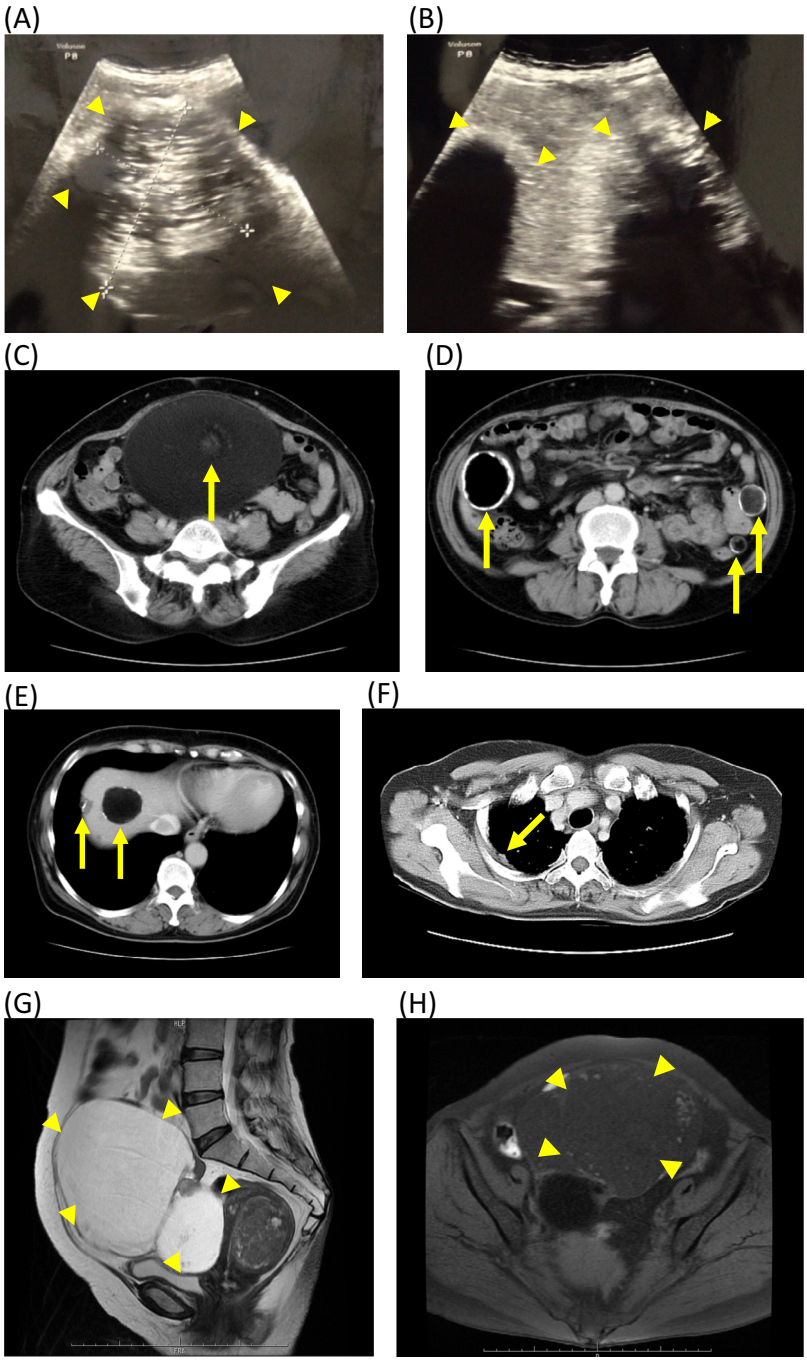

Fig. 1 – Clinical images. (A). Ultrasonography showing a large cystic mass with diffuse internal echo (arrowheads). (B). Ultrasonography showing calcified cysts with a strong acoustic shadow (arrowheads). (C). Axial contrast-enhanced CT showing a large, cystic mass containing a focal, solid, enhancing nodule component (arrow). (D). Axial contrast-enhanced CT showing multiple, variable-size, annular calcifications in the abdominal cavity (arrows). (E). Cystic lesions with distinct borders accompanied by small calcifications in the right diaphragm and the upper hepatic surface (arrows). (F). Axial contrast-enhanced CT showing a small, nodular thickening of the pleura (arrow). (G). Sagittal T2-weighted magnetic resonance imaging demonstrating a multicystic mass approximately 16 cm in diameter in the pelvic cavity (arrowheads). (H). Axial contrast-enhanced, fat-suppressed, T1-weighted magnetic resonance imaging showing a decreased fat signal (arrowheads). CT, computed tomography.

perforation in the tumor. It is believed that the ruptured contents of an MOCT, such as fat and hair, cause an inflammatory response, and chronic granulomas are formed in the peritoneal cavity. As a result of these foreign-body reactions, nodular lesions appear in various places in the abdominal cavity and sometimes in the thoracic cavity. Chronic granulomatous peritonitis is characterized by multiple white peritoneal lesions, dense adhesions, and ascites. Hence, it is

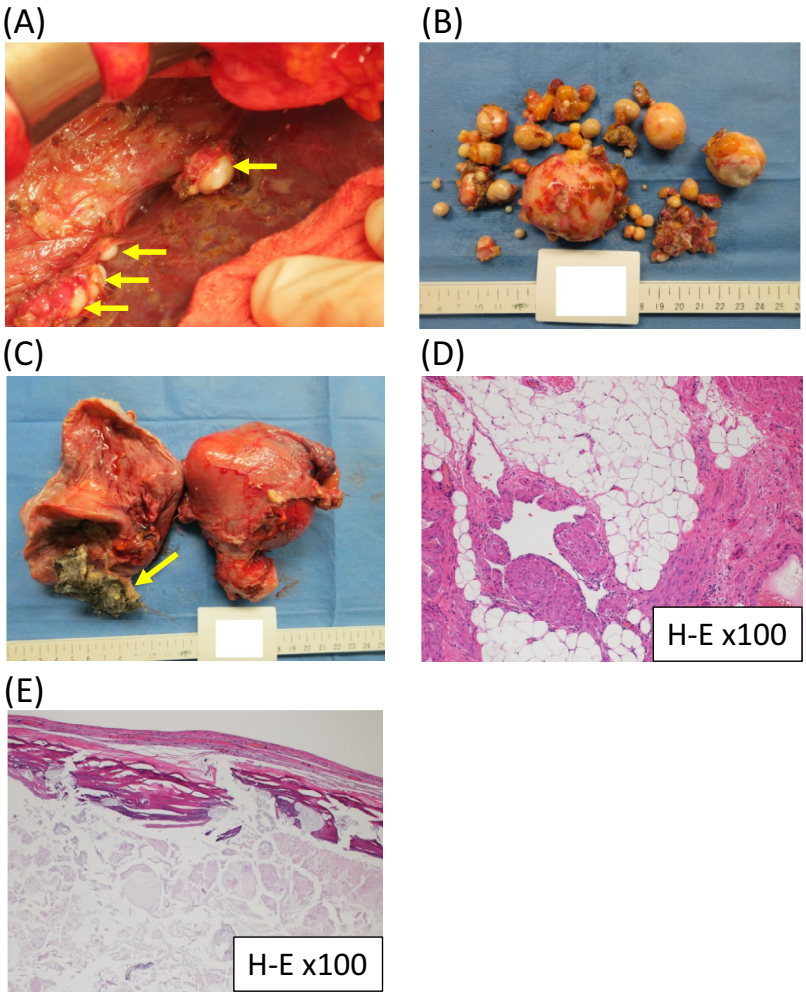

Fig. 2 – Surgical specimens and pathologic findings. (A). Intraoperative findings at exploratory laparotomy. White nodules are present along the pelvic wall (arrows). (B, C). Gross appearance of all removed white nodules (B) and the uterus with a right ovarian cyst (C). The cyst wall is smooth, and hair and adipose tissue are inside the cyst (arrow). (D, E). Histopathologic evaluation of the omentum tumors shows only mature elements (D: H-E stain, ×100). Microscopic photography of the solid components shows fibroblasts and collagen bundles indicating fibrous stroma (E: hematoxylin and eosin stain, ×100). H-E, hematoxylin and eosin.

necessary to consider the possibility of a benign mature teratoma if calcification and fibrosis of the nodules are confirmed and cellular component is poor, even when there is the appearance of multiple seeded nodules on imaging.

When multiple intraperitoneal cysts are accompanied by calcification, the differential diagnosis includes calcification of mesenteric lymph nodes, aneurysm, hydatid cyst, silicosis of the lymph nodes, free fat globules, gallstones, pancreatic stones, and a disseminated MOCT. Calcified mesenteric lymph nodes do not appear under the diaphragm, and they have a heterogeneous appearance on imaging. Our patient did not show evidence of liver dysfunction, which is typical of a hydatid cyst. She also did not display any imaging findings consistent with silicosis of the lymph nodes, free fat globules, gallstones, or pancreatic stones. The thick, white, plaque-like nodules on the visceral peritoneum observed in our patient are characteristic of chronic granulomatous peritonitis. All removed tumor masses were examined histologically; the tumor walls featured calcite with a hyaline-like structure. The primary tumor of the right ovary consisted of fat, an eosinophilic hyaline-like necrotic substance, and several mature tissues, consistent with the diagnosis of MOCT. It is likely that the MOCT contents leaked into our patient's peritoneal cavity little by little during repeated pregnancies, resulting in multiple ring calcifications.

The limitation of this report is the absence of pathologic results for the intrathoracic lesions. These might have represented dissemination of the MOCT, but there are no previous reports in which dissemination of an MOCT is pathologically proven in the pleural space.

In conclusion, we describe a rare presentation of MOCT, in which we initially strongly suspected an advanced malignancy based on the results of imaging. There is a wide spectrum of appearance of MOCT on different imaging modalities, reflecting specific pathologic or histologic components. To make an accurate diagnosis, it is necessary to understand the rare

findings associated with MOCT, as well as the common signs on different imaging modalities.

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