Ruptured Mature Cystic Teratomas Mimicking Advanced Stage Ovarian Cancer: A Report of 2 Cases Study

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Spontaneous rupture of mature cystic teratoma is uncommon. Chronic rupture of teratomas may result in granulomatous peritonitis. In rare cases, the clinical presentation and the intraoperative findings may mimic those of advanced stage ovarian cancers. Two cases of ruptured mature cystic teratomas mimicking malignancy are reported in 50- and 53-year-old patients. Both presented with abdominal distension and weight loss. Elevation of serum CA-125 (233 unit/ml) was detected in one case. Intraoperatively, nodular thickening of the omentum with marked peritoneal adhesion was observed in both cases. Ascites of 1500 ml was present in one case. Pathologic examination in both cases showed ovarian mature cystic teratomas with peritoneal granulomatous inflammations response to the released tumor content. Both patients were followed for 44 and 12 months and were free of symptoms without additional treatment other than surgery. Intraoperative pathologic consultation can help confirm the benign diagnosis and unnecessary major operation for malignancy can be avoided.

Keywords : *Ovarian tumor, Mature cystic teratoma, Spontaneous rupture, Granulomatous peritonitis, CA 125*

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Mature cystic teratoma or dermoid cyst is the most common subtype of ovarian tumors[1]. The tumors are benign germ cell tumors and mostly present with asymptomatic adnexal masses[2,3]. Spontaneous rupture of a tumor is an uncommon complication[3-6]. In rare cases, the clinical presentation and intraoperative findings of ruptured mature cystic teratomas may closely mimic malignant ovarian tumors of advanced stage[6,7]. The authors describe two cases of this unusual condition which, to our knowledge, are the first report in Thailand.

Case Report

Case 1

A 50-year-old perimenopausal Thai woman (para 1-0-0-1) was referred from a provincial hospital with a clinical diagnosis of possible ovarian cancer. She had developed abdominal discomfort and distension for the last 2 months with a 7-kg weight loss. The pelvic examination revealed an ill-defined mass of 20-cm diameter fixed to the pelvic structures. Tranvaginal ultrasonogram could not identify a definite mass in the pelvis, however, both ovaries were not well visualized. Abdominal computed tomograpgy (CT) revealed a 10-cm solid-cystic mass containing a large locule with low-density content and tooth-like calcification. Diffuse infiltrative lesion around the tumor with omental involvement was observed. The radiologic impression was a malignant teratoma with peritoneal spread. Exploratory laparotomy was performed. The laparotomy revealed a 10-cm left ovarian mass with marked surface adhesion to all pelvic structures and pelvic peritoneum. A small amount of ascites was present. The omentum with nodular mass-like thickening measured 20 cm in width and 1 cm in thickness. The left ovarian tumor was cystic and contained greasy material admixed with hair. The right ovary was not enlarged. Due to a high clinical suspicion of malignant

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tumor associated with mature teratoma, a complete surgical staging operation was performed including hysterectomy and bilateral salpingo-oophorectomy, partial omentectomy, peritoneal washing and biopsy. Except for a portion of omental lesion that was not feasible to excise, the residual peritoneal lesions were minimal. The patient was followed for 44 months postoperatively. She was well and free of symptoms without further specific treatment.

Case 2

A 53-year-old postmenopausal Thai woman (para 4-0-0-4) was referred from a private hospital with a clinical diagnosis of ovarian cancer. Two months before, she had a prolonged fever with abdominal discomfort. Malarial infection was diagnosed and she was treated accordingly. The fever subsided but the abdominal symptom persisted. In the last month, she had an increase of abdominal discomfort and distension with a 4-kg weight loss. Abdominal CT revealed bilateral ovarian masses (right 9 cm and left 3 cm) with low density and small calcification, consistent with dermoid cysts. Moderate to marked ascites was observed. There was a diffuse infiltrative lesion involving the omentum and mesentery, which could be either malignant infiltration or inflammatory process. Physical examination at Maharaj Nakorn Chiang Mai Hospital revealed marked abdominal distension due to ascites which resulted in unsatisfactory pelvic examination. The serum CA-125 level was 233 unit/ml (normal < 35 unit/ml). There was neither a history of tuberculous contact nor clinical evidence of tuberculosis. As a malignant gynecologic neoplasm could not be excluded, exploratory laparotomy was performed. The intraoperative findings included 1,500 ml of ascites, omental cake-like thickening (30 cm in width and 2-3 cm in thickness) with marked adhesion on small intestine, and bilateral ovarian masses (right 3 cm and left 9 cm) (Fig. 1). Both ovarian tumors were unilocular and contained greasy gray tan material admixed with hair. A 3 mm perforation was seen in the left ovarian tumor. The omental lesion was composed of diffuse yellowish gray nodular plaques on the omental surface and yellowish gray foci within the omental fat. Intraoperative frozen section on the omental tissue revealed granulomatous inflammatory process consistent with foreign body reaction. In addition, rare hair shafts entrapped within the omentum were further identified in the operative field. Bilateral salingo-oophorectomy and partial omentectomy with lysis of adhesion was performed. The patient was well and free of symptoms without further specific treatment 12-months postoperatively

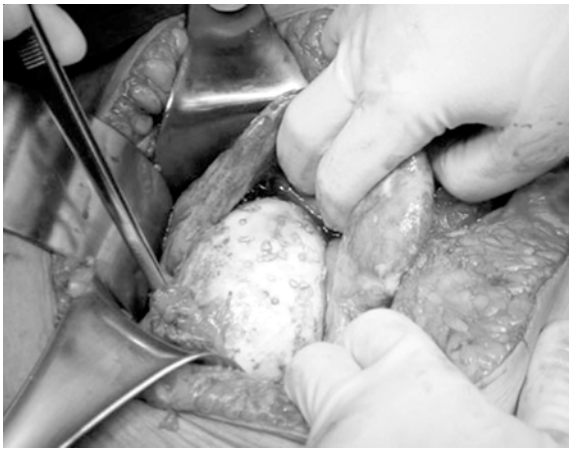

Fig. 1 Intraoperative findings at exploratory laparotomy. The omental mass is retracted to the right to show the left ovarian mass. Multiple small nodules are present on the external surface of the ovary.

Pathologic findings

On gross examination, the ovarian tumors in both cases were unilocular. The external surface of the tumor in case 1 was surrounded by dense fibrous adhesion. The external surface of the left ovarian tumor in case 2 showed multiple discrete small plaques and nodules of less than 1 cm in diameter. The tumor inner surface in both cases was smooth. Rokitansky protuberances were seen but there was no grossly malignant solid tissue. A few teeth were seen in the cyst wall of case 1.

Histologically, the cystic spaces were lined by squamous epithelium with cutaneous adnexal structures in both cases. Minor components of fat tissue, respiratory epithelium, and thyroid tissue were observed. The omental and peritoneal lesions in case 1 were characterized by granulomatous collections of epithelioid histiocytes, mostly around keratin and hair shafts. The granulomas were surrounded by dense mature fibrosis (Fig. 2). Occasional foreign-body type multinucleated giant cells were seen. The omental lesion in case 2 showed similar granulomatous inflammation with a lesser degree of fibrosis.

Discussion

Spontaneous rupture is an uncommon complication of mature cystic teratomas occurring in only 0.3-2% of cases[2-5]. Spontaneous rupture of mature cystic teratomas may be caused by adhesion of tumor

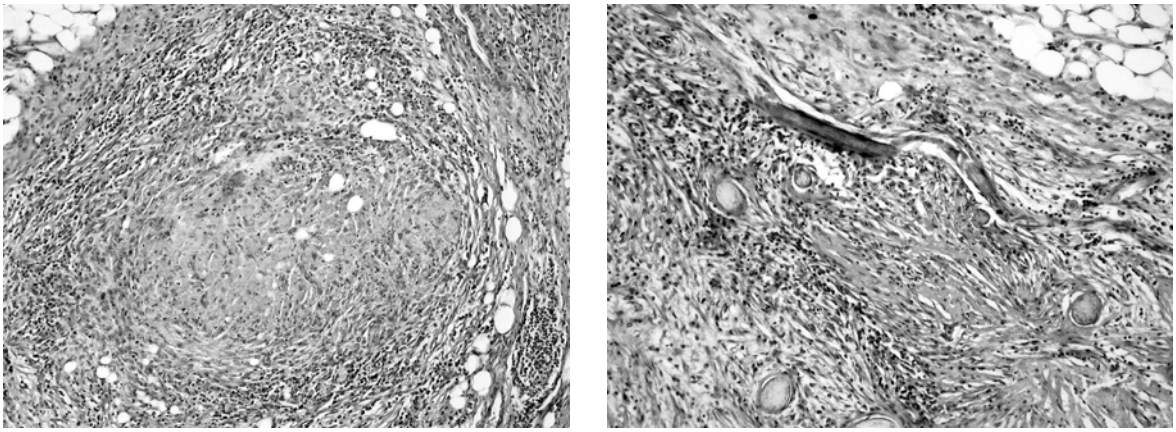

Fig. 2 Histopathologic findings in the omentum. (A) An epithelioid granuloma is surrounded by fibrotic reaction with lymphocytic infiltration (H&E stain, x100). (B) Hair shafts are enclosed by dense fibrosis (H&E stain, x100).

to adjacent structure resulting in a circulatory disturbance and subsequent necrosis of the cyst wall or by mechanical factor such as increased intraabdominal pressure upon impacted tumors or by infection of tumor[5].Mature cystic teratomas may rupture into the peritoneal cavities or, less frequently, into the adjacent visceral organs[4,5]. The clinical presentation of intraperitoneal rupture of mature cystic teratomas may be divided into two categories; acute and chronic[4,5]. In sudden rupture, the entire content of the cyst is expelled into the peritoneal cavity and causes severe chemical peritonitis that may be associated with acute abdominal crisis and shock. At laparotomy, sebaceous material admixed with hair and thick intraabdominal exudate may be observed[4]. Chronic rupture is caused by slow and continuous leakage through a small perforation of the tumor. The patient may be asymptomatic in the early period but later develop granulomatous peritonitis and associated abdominal symptoms such as progressive abdominal distension and gastrointestinal disturbances[4]. In rare cases, the presentation of peritoneal granulomatous inflammation has been reported to mimic that of advanced ovarian cancer[6]. To the authors' knowledge, approximately 20 such cases have been reported in the literature[6,7]. In the previously reported cases, significant elevation of CA-125 level has not been described.

The clinical presentation of both patients including the peri- or postmenopausal age group, weight loss, abdominal distension with ascites, relatively large ovarian tumor size, the radiologic findings of omental lesion, and, in case 2, elevated serum CA-125 level were consistent with those of advanced stage ovarian cancer. As the CT findings suggested a teratomatous nature of the ovarian masses, there should be a high suspicion of malignant transformation in mature cystic teratoma[8].At laparotomy, marked peritoneal adhesion and omental mass-like appearance in both cases was also suggestive of malignancy. Although the finding of loculated sebaceous content and hair can help in recognition that the peritoneal lesion should represent a reaction to teratoma content [4], this may be difficult to detect. Only rare hair shafts were intraoperatively identified in patient 2 and none in patient 1. Intraoperative pathology consultation with a frozen section is helpful to confirm the presence of granulomatous inflammation in the peritoneal lesion and the absence of malignancy in the ovarian teratoma. Then, conservative surgical approach can be justified. Histologically, chronic rupture of mature cystic teratoma should be considered as a possible cause of granulomatous peritonitis in female patients, in addition to other infectious and non-infectious causes.

Serum CA-125 level is a useful marker for ovarian cancer. In postmenopausal patients, significant elevation of CA-125 level (> 95 unit/ml) is highly suggestive of malignancy[9]. Granulomatous peritonitis may cause significant elevation of serum CA-125 as reported in tuberculous peritonitis [10] and in the presented case of ruptured mature cystic teratoma.

The treatment for patients with ruptured mature teratomas and granulomatous peritonitis includes removal of the ovarian tumor and excision of granulomatous masses of the omentum if this is possible[4,6,7]. Large omental masses should be removed but excision of all visible peritoneal lesions or masses may not be necessary[6]. In selected cases with significant residual inflammatory masses, the use of corticosteroid may improve postoperative resolution[6]. The clinical course of the patients with ruptured mature cystic teraoma is

usually benign[6,7]. Intraabdominal recurrence of ovarian mature teratoma following rupture is extremely rare[11].

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การแตกเองของเนื้องอกรังไข่ชนิด mature cystic teratoma ที่มีลักษณะทางคลินิกเหมือนมะเร็งรังไข่: รายงานผู้ป่วย 2 ราย

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การแตกเองของเนื้องอกรังไข่ชนิด mature cystic teratoma พบได้น้อย การแตกของเนื้องอกสามารถทำให้เกิดการอักเสบชนิด granulomatous ของเยื่อบุช่องท้อง ผู้ป่วยจำนวนน้อยมากที่มีอาการนำและสิ่งตรวจพบในระหว่างผ่าตัดคล้ายคลึงกับมะเร็งรังไข่ระยะลุกลาม ได้รายงานผู้ป่วย 2 ราย ที่มีอายุ 50 และ 53 ปี ผู้ป่วยทั้งสองรายมีอาการนำได้แก่ อืดแน่นท้อง และน้ำหนักลด การตรวจเลือดพบว่าผู้ป่วยรายหนึ่งมีค่า CA 125 สูงผิดปกติ (233 unit/ml) ระหว่างการผ่าตัดตรวจพบเนื้องอกรังไข่ ร่วมกับการหนาตัวของ omentum และมีพังผืดในช่องท้องคล้ายที่พบในมะเร็งรังไข่ ผู้ป่วยรายหนึ่งมีน้ำในช่องท้องปริมาณ 1500 มล. ลักษณะทางพยาธิวิทยาของผู้ป่วยทั้งสองราย ได้แก่ mature cystic teratoma ของรังไข่ และการอักเสบชนิด granulomatous ต่อcontent ของเนื้องอก การติดตามผู้ป่วยหลังผ่าตัดเป็นเวลา 44 และ 12 เดือนตามลำดับ พบว่าผู้ป่วยไม่มีอาการผิดปกติใด ๆ การปรึกษาและส่งตรวจชิ้นเนื้อในระหว่างการผ่าตัดในรายที่ไม่แน่ใจว่าเป็นมะเร็งรังไข่หรือไม่ จะทำให้ช่วยลดการผ่าตัดที่ทำมากเกินไปได้