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Case Report

Squamous cell carcinoma arising in a partially ruptured giant mature cystic teratoma: A case report

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ABSTRACT

Mature cystic teratomas are the most common ovarian germ cell tumors and represent 70% of the benign ovarian tumors occurring in women under age 30. In less than 2% of cases, these tumors can transform into malignancies. Squamous cell carcinoma arises most frequently from these tumors (in 80% of cases). Intra-peritoneal rupture accounts for approximately 1%-2% of cystic teratoma complications with most ruptures occurring either intra-operatively during laparoscopic surgery or due to trauma. This case report describes the common presentation, imaging, and pathologic findings of a mature cystic teratoma with 2 uncommon associated complications.

Introduction

Ovarian germ cell tumors account for approximately 20% of all ovarian neoplasms and 2% of all ovarian malignancies [1–4]. Mature cystic teratomas (MCT) are the most common ovarian germ cell tumors accounting for approximately 70% of benign ovarian tumors in women under 30 years and 50% of pediatric tumors [5–7]. Although MCTs have a wide age distribution (2-80 years with mean of 32 years), only 5% occur in postmenopausal women [8]. MCTs are composed of totipotent cells developing into well-differentiated ectodermal, endodermal, and mesodermal tissue such as teeth, hair, bone, and sebaceous glands [9]. Malignant transformation has been reported to occur in 0.17%-2% of MCTs and predominantly arises in older women, that is, those in their fifth or sixth decade of life [6,10–13]. Squamous cell carcinoma (SCC) is the most common malignancy arising from MCTs accounting for approximately 80% of cases [11]. Due to its rarity, slow growth rate (1.8 mm per year), and asymptomatic nature, most malignancies are discovered incidentally on routine exam or imaging performed for other reasons [13–15]. If present, symptoms

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most commonly include increased lower abdominal distension and pain as well as abdominal or pelvic mass or swelling [6]. Symptoms most commonly develop in patients who are postmenopausal or whose tumor has ruptured or underwent a malignant transformation [7,16]. Increased tumor size has been associated with a higher progression to malignant transformation of MCTs and a worse prognosis due to more aggressive disease [17,18]. Rupture is an unusual complication of malignant MCTs occurring in approximately 1%-2% of patients with SCC MCTs [18]. Due to the increased use of laparoscopic surgery, preoperative rupture has become increasingly rare and iatrogenic intraoperative rupture has become more frequent and can lead to a poor prognosis from spillage of teratoma contents [19,20].

Case presentation

A 55-year-old postmenopausal woman presented to her gynecologist for her first routine exam in 10 years. She complained of vague abdominal fullness, early satiety, and 40 pound weight loss in the prior 8 months. Upon presentation, the patient stated, she had a 3-day long episode of postmenopausal bleeding. On physical examination, she was noted to have a large nontender palpable mass in her abdomen. A nontender right vulvar mass was also seen during her pelvic examination.

Subsequently, the patient underwent a contrast-enhanced computed tomomgraphy of the abdomen and pelvis which revealed a large, at least 17 cm, right adnexal mass demonstrating a fat/fluid level, most consistent with a mature cystic teratoma (Fig. 1A-F). Multiple droplets of fat were also noted adjacent to but separate from a dominant mass in the lower abdomen with associated ascites and fat stranding concerning for partial rupture of the mass (Fig. 1D-F). Centrally within a dominant mass, a separate circumscribed mass with various internal densities, including solid and fat components, was suspended at an interface of fat and fluid levels (Fig. 1B and C). A right Bartholin gland cyst corresponded to the vulvar mass described on physical exam (Fig. 1A). Vaginal/cervical cytological testing was found to be negative. Serum tumor marker levels were not checked at the time of the initial workup but were subsequently requested following imaging discoveries and were normal.

Surgical planning was swiftly initiated and the patient underwent a total abdominal hysterectomy, bilateral salpingo-oophorectomy, Bartholin's cyst excision, and pelvic/para-aortic lymph node dissection. The gross specimen measured 26 × 19 × 12 cm and was partially cystic with a 10 cm solid component. Histologic sections showed features of a benign dermoid cyst (Fig. 2A and B); however, a separate solid component within a dominant mass demonstrated hallmark characteristics of in situ and invasive high-grade SCC arising within a MCT of the right ovary (Fig. 2C and D). The ovarian capsule was intact, but focal ovarian surface involvement was present. In addition, there was lipogranulomatous reaction associated with the teratoma and in the fallopian tube suggestive of preoperative rupture. Extensive lymphovascular invasion was present (Fig. 2E) and a right pelvic lymph node was positive for metastatic SCC, leading to a pathologic stage of pT1c2N1b based on TNM classification. It was also confirmed that the right vulvar mass was a Bartholin gland cyst.

The postoperative course was complicated by development of a right vulvar hematoma which was evacuated operatively. The patient recovered from the hematoma excision and was discharged shortly thereafter.

In the following months, the patient underwent port placement with goals to initiate chemotherapy. However, she subsequently declined to begin her infusion sessions. The patient was referred to palliative care specialists to assess her goals of care. In the interim, she returned to the emergency department on 2 separate occasions, in a month duration, with complaints of abdominal pain and nausea. Her follow-up imaging exhibited large volume ascites and she underwent ultrasound guided paracentesis twice with immediate symptomatic relief.

As of writing of this manuscript, the patient did not commit to her chemotherapy regimen.

Discussion

MCT, or dermoid cysts, are the most common ovarian germ cell tumors accounting for approximately 70% of benign ovarian tumors in women under 30 years and 50% of pediatric tumors [5–7]. Malignant transformation of MCTs occurs later in life with median age being reported in the fifth decade of life - 1 study comparing benign and SCC MCTs reported an average of 32.7 years old and 50.8 years, respectively [4,12,21–23]. SCC arising in a MCT is uncommon, occurring in less than 2% of cases [18]. SCC is the most common malignancy to arise in MCTs, comprising approximately 80% of malignancies, followed by adenocarcinoma and melanoma [11,24].

In our case, the patient presented simultaneously with common appearance as well as 2 rare complications, making our patient unique from previously presented cases. Our patient at 55 years supports the notion that SCC arising in MCTs occurs more commonly in older patients [25]. Symptomatically most patients with MCTs that undergo malignant transformation present with findings related to the mass effect from the tumor. Our patient's presentation with abdominal fullness, early satiety and weight loss are all common findings, which can be related to tumor mass effect [7,16,21,23,26]. Less commonly reported symptoms include bowel and bladder symptoms such as voiding difficulty, lower back pain, and fever [18,23,24,27]. Additional suspicious clinical features of this uncommon condition have been described in the literature, including postmenopausal status, large size of the tumor, solid components on imaging, and capsular invasion on histopathology [28].

The role of tumor markers has not been fully established in assessment and follow-up of MCTs. In the study by Tseng and colleagues, 16 out of 24 cases of SCC arising from MCTs had an elevated SCC antigen. Levels of SCC antigen were also elevated on serial monitoring when tumors recurred [29]. The exact utility of SCC antigen levels has yet to be established for the use of SCC MCTs. Variability in SCC antigen has been equated to tumor size and studies have not defined the utility

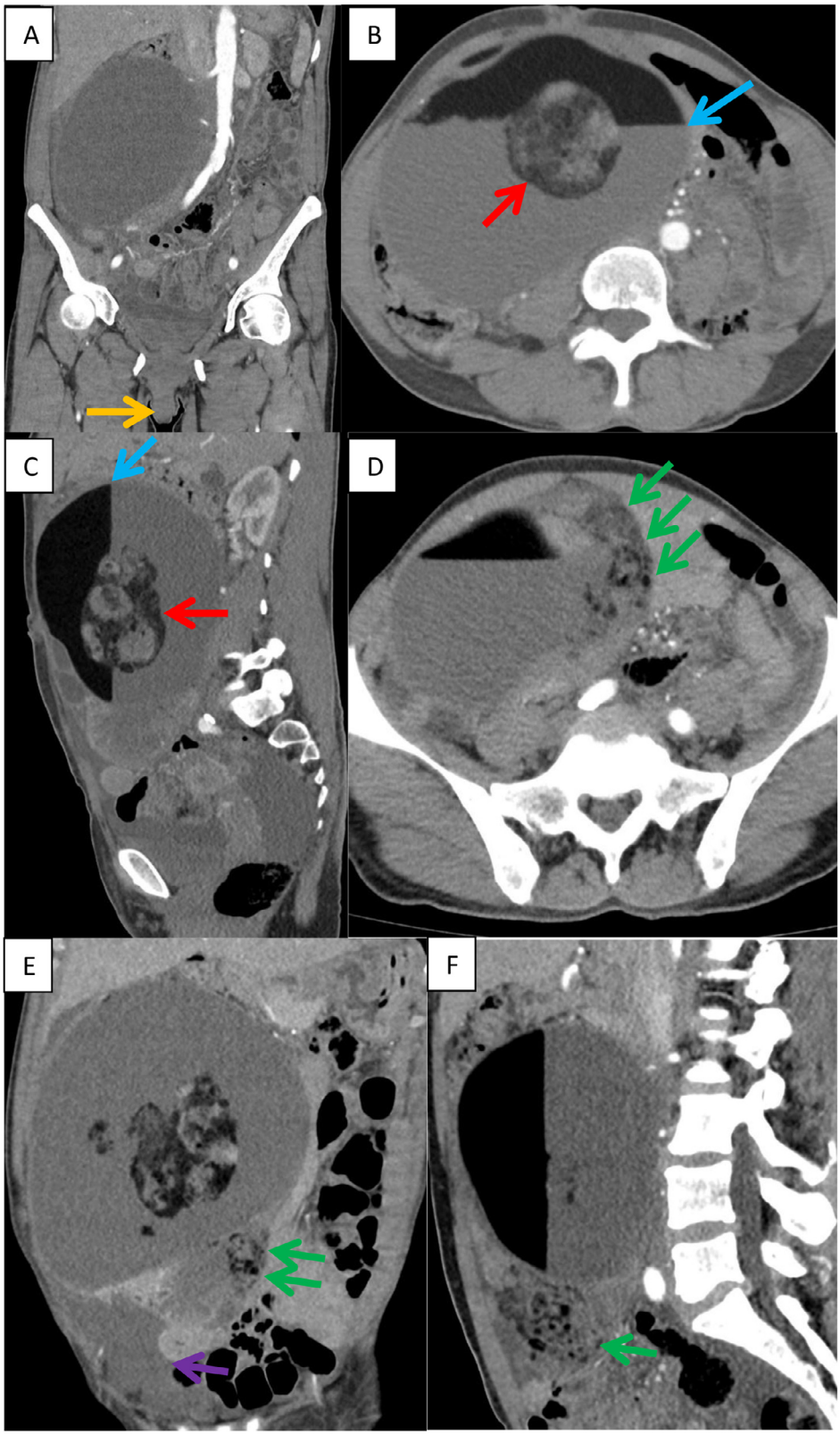

Fig. 1 – Contrast-enhanced computed tomography images: (A) through (F) – Coronal, axial, and sagittal CT images demonstrating features of MCT with internal fat/fluid level (blue arrow) and a separate smaller mass suspended within it (red arrow). Right labial Bartholin gland cyst is noted on Image A (orange arrow); (D–F) – Axial, sagittal, and coronal CT images demonstrate droplets of fat (green arrow) separate from the dominant mass with adjacent stranding and ascites (purple arrow) suggesting rupture and corresponding with lipogranulomatous reaction on histology. (Color version of this figure is available online.)

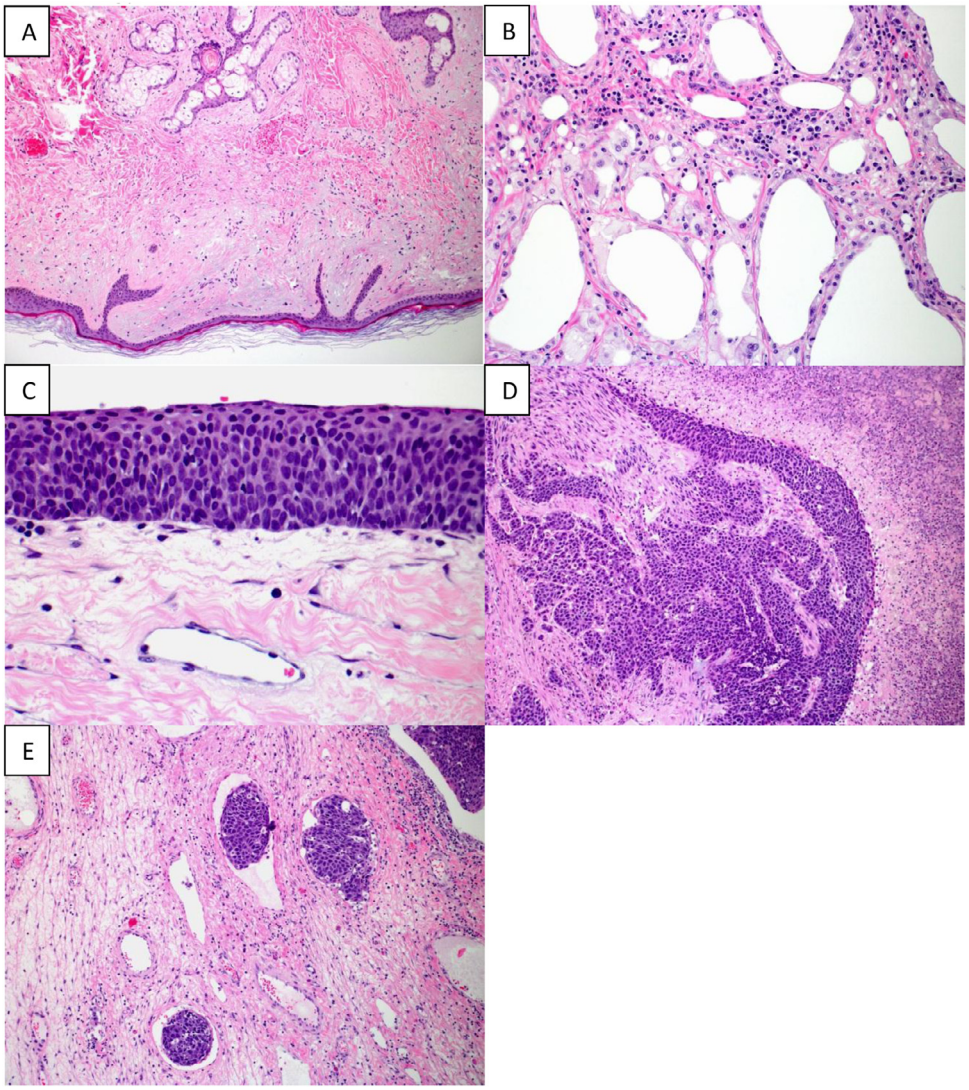

Fig. 2 – Microscopic Images: (A) – Dermoid cyst. Cyst lining with keratinized epidermis, associated adnexal structures (sebaceous glands and hair follicle), and smooth muscle (H and E, ×100); (B) – Dermoid cyst. Characteristic lipogranulomatous reaction in cyst wall (H and E, ×200); (C) – Squamous cell carcinoma in situ arising in cyst lining. There is full thickness squamous epithelial atypia characterized by enlarged, hyperchromatic nuclei and increased mitotic activity (H and E, ×400); (D) – Invasive squamous cell carcinoma with necrosis (right) (H and E, ×200); (E) – Squamous cell carcinoma with lymphovascular invasion (H and E, ×100).

of its use in early detection of early or small tumors [30,31]. Due to emergent nature of our patient's case, we did not evaluate tumor markers at the time of presentation but subsequently the markers were normal.

In prior case reports, tumors have been shown to measure up to 40 cm in largest dimension. In this case, the tumor measured up to 26 cm on gross pathology, which did not account for the fact that it had partially ruptured prior to surgical removal. A retrospective study by the Taiwanese Gynecologic Oncology Group found 52 cases of MCT with malignant transformation to SCC in Taiwanese medical centers from 1990 to 2011. Tumors measuring greater than 15 cm in diameter were found to be more aggressive than those measuring less than 15 cm [22].

Rupture accounts for approximately 1%-2% of cystic teratoma complications and potentially suggests that the original size may be larger than what was observed at presentation [32,33].

Intra-peritoneal rupture of the tumor has been described as a rare negative prognostic indicator of malignant mature cystic teratomas. In an analysis of 52 SCC arising in MCTs only 1 (1.9%) was found to have a preoperative rupture [18]. A distinct lipogranulomatous reaction in the peritoneum was demonstrated on the histopathology. Granulomatous inflammation of the peritoneum, otherwise known as lipogranuloma peritonealis occurs for various reasons, including fungal or bacterial infections, parasites, or foreign body reactions to surgery. In rare cases, a foreign body reaction may

be elicited from the ruptured contents of cystic teratomas, which is exactly what occurred in this case [34]. Few cases of intraperitoneal rupture have been described in the literature, 2 of which were described occurring due to trauma [35–37]. Of the 2 cases, 1 rupture did result in peritonitis and the authors suggested that the fat-fluid level seen on CT may suggest rupture [35], as seen in our patient. A thorough saline wash has been suggested for preoperative or perioperative ruptures to reduce complications such as chemical peritonitis, intra-abdominal adhesions, and masses [38–40]. Intraperitoneal rupture and malignant transformation of benign MCTs to SCC both occurred in the presented case despite being individually uncommon complications highlighting the uniqueness of our patient [18,41]. Due to the increasing use of laparoscopic surgery, intraoperative rupture of tumors has become more prevalent. Two cases of intraoperative rupture of SCC MCTs led to spillage of contents and dissemination of the tumor [20]. Unlike most ruptures due to laparoscopic operations, our patient's rupture occurred preoperatively, as was demonstrated on initial CT and subsequently confirmed by pathologic evaluation.

Familiarizing oneself with the presentation, imaging and microscopic features of a SCC MCT is of great significance due to the rarity and aggressive potential of the disease. As there is ongoing debate on the best management practices for this disease entity, the benefits of early diagnosis and intervention cannot be more emphasized.

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