Case Report

Frozen in Time: Intraoperative Diagnosis and Management of Malignant Transformation in Mature Cystic Teratoma

Arun Sudhakar Sanap, Medha D. Davile, Anusha A. Kamath, Shuchita R. Mundle

Department of Obstetrics and Gynecology, AIIMS, Nagpur, Maharashtra, India

Abstract

Malignant transformation (MCT) of ovary is rare complications affecting elderly, squamous cell carcinoma being the most common. The prognosis worsens with extraovarian spread. We present two cases of MCT-derived SCC. Patients exhibited abdominal lump, pain, bowel symptoms, sometimes with weight loss; imaging revealed MCT. Age (51–60), postmenopausal status, large size (>20 cm), bilaterality, and complex ovarian lesions raised suspicion of malignancy. Elevated tumor markers (e.g., cancer antigen-125 and lactate dehydrogenase) were noted in one case. Intraoperative frozen section confirmed malignancy, guiding staging laparotomy. One case was advanced stage on histopathology. Intraoperative frozen section aids optimal staging.

Keywords: *Frozen section, malignant transformation, mature cystic teratoma, postmenopausal, squamous cell carcinoma*

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Introduction

Dermoid cyst, also known as mature cystic teratoma (MCT), constitutes 10%–20% of all ovarian tumors and is the most common ovarian germ cell tumor.[1] Few cases, around 1%–2.5% of MCT undergo malignant transformation (MT).[2,3] Although MT can occur in any of the constituent tissues of teratoma, squamous cell carcinoma (SCC) is the most commonly observed in over 80% of the MTs.[4-6] We present here two case reports of postmenopausal women diagnosed with SCC transformation within an MCT, highlighting the pivotal role of intraoperative frozen section in guiding optimal treatment strategies.

Case Reports

Case 1

A 53-year-old, para 3, postmenopausal female presented with a complaint of abdominal distension for 4 months. On inquiry, the patient gave a history of loss of appetite and abdominal bloating. Clinical examination revealed a central, solid-cystic mass, corresponding to a 36-week gravid uterus, with bosselated surface in the lower abdomen with restricted mobility. A bimanual pelvic examination identified a solid-cystic mass in the pouch of Douglas, uterus could not be felt separately. Ultrasound imaging showed bilateral complex ovarian cysts with areas of calcification. A contrast-enhanced computed tomography (CT) scan delineated a large 20 cm × 15 cm × 23 cm abdominopelvic, fat-containing cystic lesion on the right side and a similar fat-containing cystic lesion measuring 7 cm × 5 cm × 6 cm on the left side suggestive of mature teratoma. Mass appeared compressing bilateral ureters. Epithelial ovarian tumor markers such as cancer antigen 125 (CA-125), carcinoembryonic antigen (CEA), and lactate dehydrogenase were raised, 75.36 U/ml, 5.73 ng/ml, and 249 U/L, respectively. Other markers including beta-human chorionic gonadotropin, alfa-fetoprotein, CA 19.9 were within normal limits. Considering the age (sixth decade), the postmenopausal status of the patient, and the large size (>20 cm), bilaterality, and complex pattern of the ovarian lesion; ovarian malignancy was suspected. Exploratory laparotomy was planned with an arrangement of a frozen section. Upon opening the abdomen by midline vertical incision, a right ovarian tumor measuring 30 cm in the largest

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Address for correspondence: Dr. Medha D. Davile, OPD 316, Department of Obstetrics and Gynecology, AIIMS, MIHAN, Nagpur - 441 108, Maharashtra, India.
E-mail: nagpure.medha@gmail.com

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dimension along with a small left ovarian cystic lesion of 8 cm adherent to the pelvic peritoneum in the pouch of Douglas was found [Figure 1]. Bilateral salpingo-oophorectomy was done. The cut section of the mass showed brownish-yellow-colored pultaceous material containing multiloculated lesions with solid areas and hair. The wall of the lesion was carefully inspected. Approximately 1–2 cm thickened and hard mass with area of hemorrhage was present in the wall of the right-sided ovarian lesion. Intraoperative frozen section analysis showed MCT on the left side and SCC within the MCT on the right side. A complete staging laparotomy, inclusive of peritoneal fluid cytology, peritoneal biopsy, infracolic omentectomy, and lymph node dissection, was performed. Peritoneal fluid cytology was negative for malignant cells. Histopathology report confirmed the diagnosis of MCT with MT (SCC), other structures were free of tumor (peritoneum, lymph nodes, and omentum), stage pT1a [Figures 2 and 3]. The postoperative CT scan showed no residual disease. Medical oncologist opined no need for chemotherapy. She was well till the follow-up of 2 years.

Case 2

In another similar case, a 53-year-old, para 2, a postmenopausal woman with hypertension and diabetes, presented with a complaint of pain in the lower abdomen bloating, recurrent gastrointestinal system upset, and loss of appetite for a month. On inquiry, the patient gave a history of weight loss evident by loosening of the clothes. On examination, a central, mobile mass, firm to hard in consistency was present in the lower abdomen, corresponding to a 24-week gravid uterus. Per speculum examination was unremarkable. Bimanual pelvic examination confirmed the findings of abdominal and speculum examination, and the uterus could be felt separately. Radiological imaging (ultrasonography and CT scan) showed a large biloculated complex solid-cystic lesion (12 cm × 11 cm and 9 cm × 8 cm) with heterogeneous soft-tissue component and calcification, and both ovaries were not seen separately from the lesion with peritoneal lipomatosis. The tumor markers such as CA-125, CEA, and CA 19.9 were within normal limits. Exploratory laparotomy with midline vertical incision showed a large 20 cm × 15 cm × 15 cm right ovarian solid cystic lesion adhered to bowel and bladder. The perineum showed a nodular appearance at multiple sites. The omentum was thickened, nodular, and adherent to the ovarian lesion. The right salpingo-oophorectomy was done; the cut section showed a solid-cystic lesion with brownish-colored pultaceous material containing multiloculated lesions with solid areas and hair. The intraoperative frozen section revealed SCC within the MCT. A complete staging laparotomy was done. Final histopathology confirmed the diagnosis and also revealed omental metastasis. Postoperatively, platinum-based chemotherapy was started but the patient succumbed during chemotherapy.

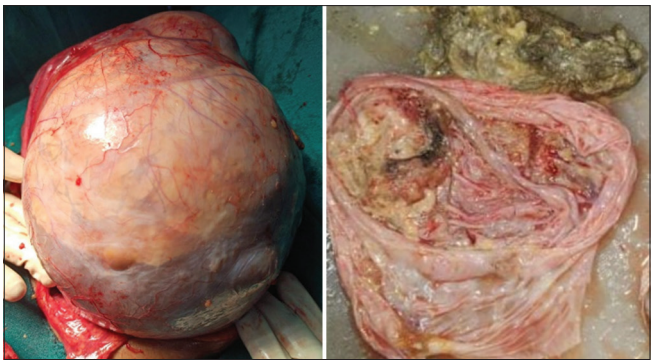

Figure 1: Intraoperative picture and postoperative cut open specimen showing pultaceous material and hairs

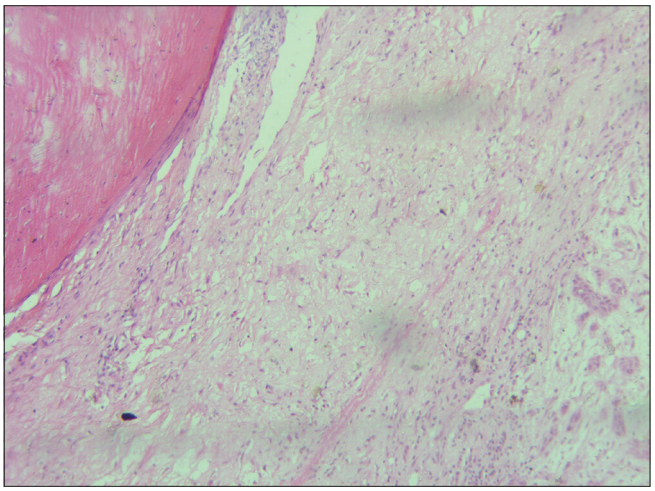

Figure 2: Bone tissue component of mature cystic teratoma along with carcinoma cells

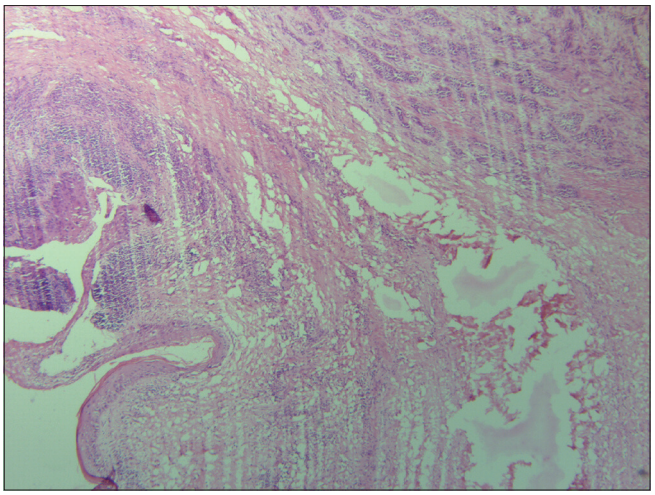

Figure 3: Lymphoid tissue component of mature cystic teratoma along with carcinoma cells

Discussion

MCT, characterized by well-differentiated germ cell elements representing all three layers, rarely undergoes MT. Factors such as prolonged exposure to pelvic carcinogens and potential association with high-risk human papillomavirus infections have been implicated in the malignant change.[6,7]

The MT of MCT is more common in postmenopausal patients with a mean age of around 50 years.[2] In this case report, both the patients were postmenopausal and aged 51 and 60 years. Clinical diagnosis of MT is challenging with nonspecific symptoms and signs. There is a significant difference in the size of MCT and MCT with MT reported in various studies with later being the larger Upto 11-20 cm in size.[1,2,4,8] In the present case reports, the size of MCT was >20 cm in both patients.

Radiologically, MCT may manifest as contrast-enhanced solid components and transmural extension. Ultrasound is the method of choice in the diagnosis of MCT. Magnetic resonance imaging and computerized tomography scan of the pelvic-abdominal region better visualize the fatty and sebaceous contents of the MCT. Unfortunately, radiological imaging fails to diagnose the MT except when there is advanced cancer with the ovarian capsule breech and local spread. Adhesion to neighboring structures, presence of nodules, increased wall thickness, and presence of necrosis and hemorrhage raise the suspicion of malignancy.[6,9]

Tumor markers such as CA-125, CEA, and CA 19.9 are often elevated in malignant teratomas but lack specificity.[10] In one of our cases, CA-125, CEA, and CA 19.9 were within normal limits, underscoring the challenge of relying solely on tumor markers for diagnosis. Notably, squamous cell carcinoma (SCC) antigen, while questioned as a reliable marker, could offer utility in cases of recurrence.[6,11] The SCC antigen testing was not done in our case.

Intraoperative frozen section in suspected cases of MCT plays a vital role in the surgical management of the patient and avoiding second surgery in these cases. In our cases, complete staging was done based on the frozen section report.

The prognosis for pure SCC arising in the ovary is notoriously poor when compared to other epithelial ovarian malignancies.[12] Survival rates and responses to chemotherapy in SCC arising in the context of MCT or endometriosis may differ, suggesting potential variations in chemotherapy sensitivity.[13] However, the lack of a consensus regarding adjuvant treatment complicates the management of MCT with MT.

There is ongoing uncertainty surrounding the efficacy of chemotherapy or radiation in MT cases of MCT. Some studies advocate for combination chemotherapy, employing agents such as paclitaxel and carboplatin, reporting extended progression-free intervals.[14] Yet, the broader applicability of such approaches remains unclear, necessitating further investigation into standardized adjuvant treatments.

In conclusion, although MT of MCT is a rare occurrence, its consideration is crucial, particularly in postmenopausal women presenting with sizable tumors. The nonspecific nature of radiological findings and tumor markers underscores the importance of intraoperative frozen sections for definitive diagnosis and comprehensive staging. These tumors typically carry a poor prognosis, emphasizing the need for further research to clarify the role of chemotherapy or radiation in their management. Platinum-based chemotherapy remains a common recommendation, but a consensus on adjuvant treatment strategies is yet to be established and more studies are required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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