ADENO-SQUAMOUS CARCINOMA ARISING IN MATURE CYSTIC TERATOMA: A RARE CASE REPORT

Shalini Shukla¹, Sanjay Nigam², Jayant Rai³, Parul Joshi⁴, Neha Ahuja⁵

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ABSTRACT: The most frequent ovarian germ cell tumours are mature cystic teratoma (MCTs), comprising 10-25% of all ovarian neoplasms. Malignant transformation in a mature cystic teratoma of the ovary is a rare complication. Squamous cell carcinoma is the most common transformation followed by adenocarcinoma. **CASE REPORT:** A 36 year old female presented with abdominal pain, fullness, difficulty in periods since 6 months. According to examination and CECT abdomen 13.5x13cm mass is found on the left adnexal region. Patient underwent oophrectomy. According to histopathological diagnosis, adenosquamous carcinoma arising in mature cystic teratoma is diagnosed as a reason for the mass in the left adnexa of the patient.

KEYWORDS: Ovary, Teratoma, Carcinoma, CECT, Adnexa.

INTRODUCTION: The most frequent ovarian germ cell tumors are mature cystic teratomas (MCT_s), comprising 10–25% of all ovarian neoplasms and 5% of ovarian cancers.^[1,2] It is believed that they arise from postmeiotic germ cells,^[3]consisting of all three germ-cell layers(ectoderm, mesoderm, and endoderm)^[2] MCTs grow in the fifth to sixth decade of a woman's life,^[4] nevertheless they are also very common in women of childbearing age, found in both ovaries in 10–17% of patients.^[5] Their clinical presentation seems to be similar to all ovarian tumors as they cause abdominal pain, constipation, bleeding, weight loss, urinary frequency and fever.^[1,3]The potential of undergoing malignant transformation(one or more of the three different mature elements of MCTs) is present, typically in postmenopausal women, with a frequency of 0.17–3%.^[6,7] Most of MCTs are detected 10–15 years before secondary malignant transformation possibly as a result of exposure in different pelvic carcinogens which trigger malignant changes in mature tissue.^[8]. Due to the high density of ectoderm in these tumors, not surprisingly the most common malignant tumor arising from them is squamous cell carcinoma(SCC),^[2,7] while various adenocarcinomas, carcinoid tumors, melanomas and various soft tissue sarcomas have also been reported.^[2,4,9]

CASE REPORT: A 36 years old female presented with the complaints of abdominal pain, abdominal fullness and difficulty in periods since 6 months. She does not have a known medical history and family history of cancer. Physical examination of lower abdomen revealed fullness and feeling of a mass. CECT abdomen showed a well-defined cystic lesion of size 13.5x13cm in left adnexal region extending upto umbilicus. A heterogeneously enhancing nodule of sized 3x2.5cm seen in anterior aspect of lesion. Comple blood count, Liver function, Kidney function test values were normal.

Tumour markers CA-125, CA 19.9, CEA were normal too. Patient underwent oophorectomy. Grossly wereceived ovarian cyst measuring 13x11x10cm.Cut surface shows multiple cyst like spaces filled with cheesy material and bunch of hair. (Fig. 1, 2) Wall is thickened at places (Fig. 2 indicated by arrow) and shows grayish white firm area measuring 0.8x0.5cm.Microscopically sections from the

thickened area show features of adenosquamous carcinoma arising in mature cystic teratoma. (Fig. 3,4,5,6)

DISCUSSION: Malignant transformation of MCTs may arise from any of three germ cell layers present in the teratoma, with an average frequency of 1-2%.^[9] Age and size are predictors of malignant transformation in benign teratoma. It is of great importance to know that MCTs arising in patients older than 45 years old sustain a higher suspicion of malignancy.^[10] Malignant transformed mature cystic teratomas have a larger size (Mean size of 15 cm) compared with MCTs (mean size of 6–9 cm).^[11]

Symptoms at presentation are variable in both diagnoses with some patients presenting with acute abdominal pain and others with constitutional symptoms such as fatigue, urinary symptoms and anorexia/weight loss.^[12] In our case, woman's age was 36 years old and presented with the complains of abdominal pain and it is worth to mention her premenopausal status. Although tumor markers may be raised in patients with squamous cell carcinoma arising from MCT, it is difficult to use tumor markers to distinguish between MCT and squamous cell carcinoma arising from an MCT since tumor markers can also be elevated with MCT.^[2]

For example, squamous cell antigen (SCC) levels were found to be significantly higher in patients with squamous cell carcinoma arising from MCT than with MCT alone. However, mean levels in squamous cell carcinomas are lower than in patients with adenocarcinomas, and cannot predict the diagnosis preoperatively.^[13] CA19.9 is another tumor marker found to be significantly higher in patients with squamous cell carcinoma arising from MCT than with MCT alone, however it is also a difficult marker to use in preoperative screening since the mean levels of CA 19.9 are found to be elevated in patients with MCT alone.^[13]

In our case all tumour markers were in normal range. Radiologically, mature teratomas may demonstrate a broad spectrum of findings ranging from a purely cystic mass, a fat-containing mass or a heterogeneous soft tissue mass. Classic diagnostic findings for MCT include fat attenuation within a cyst that possibly also contains calcification.^[14] Imaging features concerning for malignant transformation include thick walls, enhancing solid components or papillary projections within the cyst, peritoneal deposits or lymphadenopathy. In our case CECT abdomen showed a well-defined cystic lesion of size 13.5x13cm with a heterogeneously enhancing nodule of sized 3x2.5cm seen in anterior aspect of lesion.

As a result of the rarity of adenoaquamous carcinomaarising in MCT, there is no standard treatment and most often patients are treated in the same way with those patientsas with epithelial ovarian cancer.^[2] Multiple case series recommend surgery including total hysterectomy, bilateral salpingo-oophorectomy, omentectomy and pelvic-paraortic lymph node dissection with further platinum-based agent chemotherapy.^[2,12] The role of radiotherapy still remains unclear.^[2,12] In our case, after the final pathologic specimen result, patient proceeded to a second surgery including total hysterectomy, left salpingo-oophorectomy, omentectomy and pelvic node dissection.

Fig. 1: Gross specimen of ovarian cyst.

Fig. 2: Cut Surface shows cheesy material with bunch of hair and thickened wall indicated byBlack arrow.



Fig. 3: Malignant squamous epithelium.

Fig. 4: Keratin pearl (400X) with keratin pearls (40X).



Fig. 5: Tumour cells arranged.

Fig. 6: Tumour cells showing pleomorphism in glandular pattern (40X) with anisokaryosis & prominent nucleoli (400X).



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CONCLUSION: A rare and unusual disorder is adenosquamous carcinomaarising in an MCT. As an exceedingly rare disease there is no standard of therapy, with most cases progressing to surgery followed by chemotherapy.

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AUTHORS:

- 1. Shalini Shukla
- 2. Sanjay Nigam
- 3. Jayant Rai
- 4. Parul Joshi
- 5. Neha Ahuja

PARTICULARS OF CONTRIBUTORS:

- 1. Lecturer, Department of Pathology, GSVM Medical College, Kanpur.
- 2. Professor, Department of Pathology, Rama Medical College and Research Centre, Kanpur.
- 3. 2nd Year Junior Resident, Department of Pharmacology, GMC, Surat.

FINANCIAL OR OTHER COMPETING INTERESTS: None

- 4. Assistant Professor, Department of Pathology, Rama Medical College and Research Centre, Kanpur.
- 5. Consultant Pathologist, Paliwal Diagnostic Private Limited.

NAME ADDRESS EMAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Sanjay Nigam, Professor of Pathology, Rama Medical College and Research Centre, Kanpur. E-mail: sknigam@yahoo.com

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