

## Squamous Cell Ovarian Cancer on Mature Cystic Teratoma Base: A Rare Case and Literature Review

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### Abstract

Squamous cell carcinoma (SCC) is the most common type, that is 80-90%, of malignant tumor developing in benign cystic teratoma. Others are adenocarcinoma, undifferentiated sarcoma and carcinoma, and papillary carcinoma. Squamous cell carcinoma arising from mature cystic teratoma is a rare pathologic event accounting for only 1-2% of cases. Primary squamous cell carcinoma of ovaries is very rarely seen in malignancies of ovarian structures. Most of the cases develop either on mature cystic teratoma, Brenner's tumor or endometriosis. In this case presentation, we present a 40-year old female patient with unilateral ovarian squamous cell cancer on mature cystic teratoma base during her reproductive term and we also make a literature review about the subject.

**Keywords:** Ovary; Mature cystic teratoma; Squamous cell carcinoma

### Introduction

Mature cystic teratomas (MCT), also called dermoid cysts, is the most commonly seen germ-cell tumor of ovaries. They make up about 10-20% of all ovarian tumors during reproductive term [1]. Usually they are observed during reproductive period and characterized by abdominal and groin pain, however asymptomatic cases are also present [2]. Malignancy transformation is seen in about 1-4% cases with more commonly in post-menopausal women and out of these malignancies, 75-80% of them are squamous cell cancers [1,3]. When literature is reviewed, there are also malignant transformations into adenocarcinoma, melanoma, carcinoid tumor, small cell carcinoma and sarcomas [2,4]. MCT's with malignant properties usually spread out to close organs through local invasion and intraperitoneal seeding, which causes symptoms associated with affected organs [4,5]. In this case report, we present a 40-year old female patient with unilateral ovarian squamous cell cancer on mature cystic teratoma base during her reproductive term and we also make a literature review about the subject.

### Case Presentation

A 40-year old multigravida patient in her fertile period presented to our clinic with abdominal pain ongoing for 7 months. In pelvic examination, a gross mass lesion that is echogenic which contains solid-cystic areas and about 90 × 60 mm in size, filling right adnexial area was detected. Lab results were within normal ranges with the exception of CA125 (62 U/ml). Patient underwent laparotomy through an median lower abdominal incision and a cystic mass with regular contours and about 80 × 70 in diameter, originating from right ovary was inspected. During intraoperative period, no other pathologies were detected apart from right ovarian mass, left ovary was visually inspected and palpated and peritoneal fluid samples in addition to biopsies from peritoneum and omentum were taken from the patient. Due to a strong wish to keep her fertility intact, the patient had right salpingo-oophorectomy and the excised mass was sent for speedy pathology inspection (Figure 1). The result was reported as benign. Patient developed no post-operative complications and the pathology examination of the patient confirmed our squamous carcinoma developed on teratoma base. Microscopically, keratinising squamous epithelium with underlying skin adnexa, respiratory type epithelium, cartilage and neural tissue were identified. Focally, the cyst lining consisted of non-keratinising squamous epithelium with marked acanthosis and atypia and superficially SCC.



Figure 1: Macroscopic view of sebaceous and hair material.

The lesion measured 1 cm. The external surface of the cyst was intact (Figures 2A and 2B). Patient was followed-up in our clinic later (Figure 3).

### Discussion

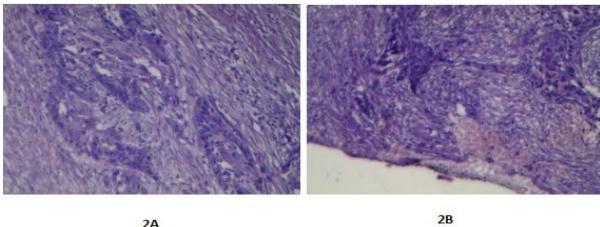
Mature cystic teratoma contains 3 ectoderm leaves. Out of these ectodermal tissues, skin is usually seen within dermoid cysts [6,7]. In mature cystic teratomas are common tumors which make up about 10-25% of all ovarian neoplasms and 60% of all benign ovary neoplasms [8]. Although they are usually asymptomatic, a small number of patients can show non-specific symptoms such as abdominal pain and bloating. Dermoid cyst has a relatively slow-growth speed in most of the cases and does not give out many clinical symptoms before growing into a very large size or development of complications such as torsion

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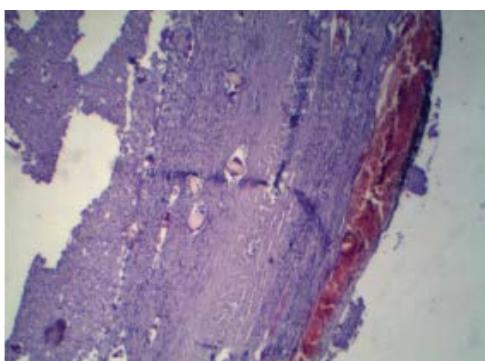
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**Figure 2: (A and B)** Malignant proliferation seen in histopathological examination (H&E stain under 20x magnification).



**Figure 3:** Benign cyst wall including hair shafts in histopathological examination (H&E stain under 20x magnification).

and rupture of the cyst [9]. MCTs have a typical macroscopic view and they contain uniloculated sebaceous matter in about 88% of the cases. Hair, skin, muscle and other tissues can be observed within tumor. A solid part which creeps towards within the mass and called Rokitansky nodule with a characteristic ultrasonographic (USG) view can be found in large numbers or none at all within the tumor [10]. Microscopic examination shows that ectodermal structures are more prominent and covered with keratinized squamous epithelium. Malignancy transformation rates were reported differently in different studies, but they are thought to be 1-2% in all cases. The most common malignancy transformation is squamous cell cancer [11]. Squamous cell cancer transformation rates in dermoid cyst-based malignancies are about 65-90% [9,12]. In the event of ovarian masses, malignancy must be considered until proven otherwise. Risk factors include advanced age (>45), postmenopausal period, tumor size >10 cm and increased solid components within tumor [13,14]. Interestingly enough, our case was a relatively young within her reproductive period.

It is difficult to detect teratoma cases with malignant transformations with examinations and radiological studies during preoperative term. Those cases are usually unilateral and the mass seen on USG has thick walls and possibly have more solid areas than cystic areas. Tumor sizes change between 2 and 15 cm. As mentioned, since clinical detection of malignant transformation is hard, some serum markers such as squamous cell cancer antibody (SCC) and P53 mutations are thought to be helpful in this subject in some of the previous studies [15]. However, studies on CA 125, CEA, CA19-9 and SCC antibodies are inconclusive [16,17]. In our case, tumor size was about 9 cm and CA-125 was 62U/ml.

Metastasis to surrounding organs through direct invasion is common in malignant transformation of teratoma cases. It is reported

that about in 60% of the cases, tumor crossed over ovary capsule and spread into neighboring tissues [18]. Furthermore, it could lead to acid build-up within peritoneal cavity. In our case, no acid was detected within peritoneal cavity and the tumor did not cross ovarian capsule yet. Left ovary and tube of the patient had a completely normal appearance yet right ovary was completely damaged by tumor tissue. Considering the facts that the left ovary had an healthy appearance, right ovary had no savable tissue left and speedy pathology result was uncertain, salphingo-oophorectomy was performed. The paraffin-block examination, which proved our malignancy diagnosis, also justifies the surgical intervention. Due to suspected views of the solid and hemorrhagic areas of the mass, taking samples of peritoneal lavage fluid samples in addition to multiple peritoneal biopsies is a correct decision.

Independent of etiology, most primary ovarian squamous cell cancer cases were advised radical surgery (TAH-BSO/Total Abdominal Hysterectomy – Bilateral Salphingo-oophorectomy) and additional adjuvant therapies [19]. However, in some of the publications, using adjuvant therapies is still debated. In young and fertile patients with a desire to keep their fertility intact, USO, surgical grading and close follow-up are advised [20]. In our case, due to fertility concern of the patient, we performed unilateral salphingo-oophorectomy and the patient was followed up closely during post-operative period.

Tseng et al. [21] reported 100% of 2-year disease-specific survival rate in 4 Grade IA patients without adjuvant therapy. In surgical treatment, Hackethal et al. [22] reported that additional lymphadenectomy increases survival rates (59.2 vs 40.4 months respectively), however, omentectomy has no effect on these rates.

In some case reports, concerning adjuvant therapies following radical surgical treatment in advanced cases, early paclitaxel addition to platinum agents are discussed [23]. In a study, a Grade 2B-3C patient group received cisplatin based antineoplastic chemotherapy with and without radiotherapy [21]. The role of radiotherapy in treatment is still controversial. In a limited number of case reports, radiotherapy is reported to have a limited effect on treatment [19]. On the contrary, Dos Santos et al. [1] reported that squamous cell cancers (SCC) are radio-sensitive tumors and suggested complete pelvis radiotherapy in addition to platinum chemotherapy in Grade 1-2 patients.

Unfortunately, the expected prognosis is bad in SCC's developed on mature cystic teratoma bases. Many cases with advanced stage tumors by malignant transformation into squamous cell carcinoma (one study reports them to be around 80%) die within months of diagnosis [19].

In a study, 5-year disease-specific survival rates were reported to be 95% in Stage 1, 80% in Stage 2 and 0% in Stages 3-4 [3]. In another study, the same value was reported as 100% in Stage 2 and 30% for Stage 3, but 0% for Stage 4 [21]. On the other hand, in squamous cell cancer transformations developing on dermoid cyst base usually have a relatively better prognosis in comparison with transformations on endometriosis base, but still, the basic factor that affects the prognosis is grade and stage of tumors [24].

## Conclusion

To sum up, patients with mature cystic teratoma, especially if they are over 40, must be closely examined for malignant transformations. To check the existence of malignant mass properties, ultrasonography, gynecological exam and lab results can be used and if the patient is in advanced age, a detailed histopathological examination is imperative in order not to miss an accompanying malignancy.

## Declarations

### Authors contributions

OB assembled, analyzed and interpreted the patient data. All authors contributed to writing the manuscript. All authors read and approved the final manuscript.

### Ethics approval and consent to participate

Not applicable

### Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

### Competing interests

The authors declare that they have no competing interests.

### Availability of data and material

The authors agree to make the raw data and materials described in our manuscript freely available.

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