

## Case Report

# Squamous Cell Carcinoma Arising in Mature Teratoma of the Ovary Masquerading as Abdominal Tuberculosis

Shreeya Taresh Indulkar, Manisha S. Khare, Vinaya B. Shah, Archana L. Khade

Department of Pathology,  
HBT Medical College and  
Dr. R. N. Cooper Hospital,  
Mumbai, Maharashtra, India

### ABSTRACT

Pure squamous cell carcinoma (SCC) of the ovary is rare. SCC can arise in a mature teratoma (MT), ovarian endometriosis or in a Brenner tumor. SCC is the most common malignant transformation arising in MT and comprises 80% of all cases. Such neoplastic transformations are extremely difficult either to predict or detect early. The mechanism of malignant transformation has not been completely understood. Due to the rarity and the aggressive course, diagnosis and treatment constitute a big challenge. We report a case of SCC arising in MT presenting with a huge abdominopelvic mass and abundant peritoneal collections clinically masquerading as abdominal tuberculosis. A review of literature with special emphasis on prognosis and treatment modalities is also presented.

**KEYWORDS:** Malignant transformation, mature cystic teratoma, ovary, squamous cell carcinoma

## INTRODUCTION

Malignant transformation of mature teratoma (MT) is rare and can arise from any component of teratoma; however, squamous cell carcinoma (SCC) is the most common malignancy and comprises 80% of all malignant transformations.<sup>[1]</sup> The mechanism of malignant transformation has not been completely understood. It is often challenging to diagnose an ovarian SCC arising in a teratoma preoperatively, due to its rarity, vague symptoms, and aggressive course.

## CASE REPORT

A 40-year-old female presented with acute abdominal pain and dysmenorrhea for the past 2 days. Per-abdomen examination showed a large firm abdominal mass. Her hemogram revealed no abnormality, except anemia. Serum tumor markers, i.e., coelomic antigen (CA) 125, alpha-fetoprotein, and beta-human chorionic gonadotropin, were normal. Computed tomography (CT) scan showed large multiloculated collections encasing the uterus and bowel, extending to the supraumbilical region. A separate soft-tissue left pelvic mass measuring 6.8 cm × 7.4 cm × 6.9 cm was seen. The right ovary was not well appreciated and was possibly compressed [Figure 1a].

The patient had similar complaints 2 years back. Ultrasonography of the abdomen showed large abdominal collections and generalized omental thickening, suspicious of abdominal Koch's. Abdominopelvic CT, in addition, revealed bilateral bulky ovaries. Right ovary measured 3.9 cm × 3.2 cm and showed enhancing solid component, suspicious of ovarian malignancy. Diagnostic laparoscopy revealed abundant white material filling the abdomen mimicking caseous material, which on mycobacterial culture showed no growth. The patient was empirically started on anti-Koch's treatment. The patient's condition worsened; hence, diagnostic laparotomy with right ovarian cystectomy was performed. Histopathology diagnosis was dermoid cyst.

Currently, the patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy and excision of pelvic mass. The right adnexal mass measured 10 cm × 5 cm × 4 cm. The cut surface was

**Address for correspondence:** Dr. Shreeya Taresh Indulkar,  
Department of Pathology, HBT Medical College and  
Dr. R. N. Cooper Hospital, Juhu, Mumbai - 400 056,  
Maharashtra, India.  
E-mail: shreeyaindulkar89@gmail.com

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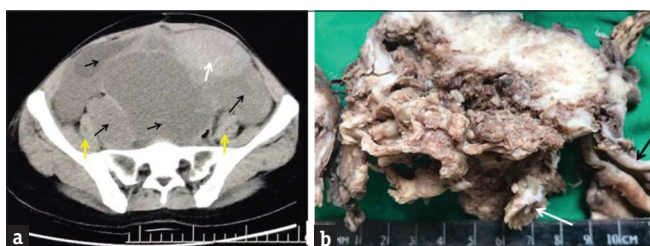
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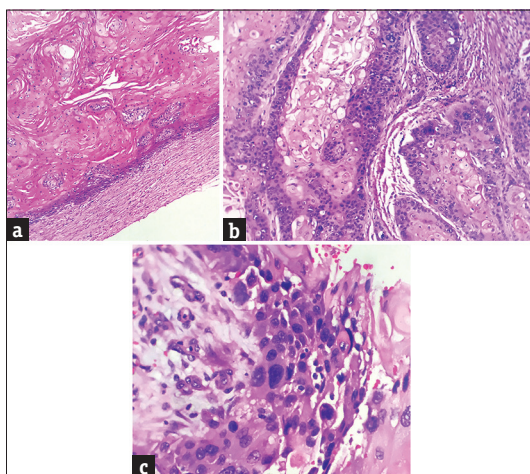
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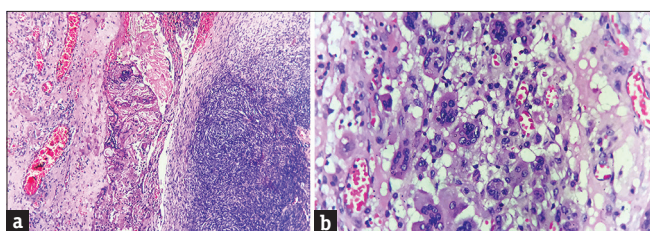
predominantly solid and was gray-white friable. The cystic part was thick walled and focally showed white pultaceous material. The pelvic mass was solid cystic, measuring 10 cm × 8 cm × 5 cm. Left ovary externally showed multiple gray-white nodules ranging from 0.2 to 0.5 cm [Figure 1b]. Histological examination showed a moderately differentiated SCC [Figure 2]. Left ovary showed keratinous material with abundant foreign body granulomatous reaction [Figure 3]. Histopathological diagnosis of ovarian SCC stage IIB (TNM - T2b N0 M0) was given. She was advised adjuvant chemotherapy and close follow-up.



**Figure 1:** (a) Computed tomography scan showing multiloculated collections (black arrows) encasing the uterus (white arrow). Bilateral ovaries are seen compressed (yellow arrows). (b) Pelvic mass showing solid cystic part (black arrow) with focal keratinous areas (white arrow)



**Figure 2:** (a) Right adnexal mass showing stratified squamous epithelium and abundant keratinous material (H and E, ×100). (b) The section from the pelvic mass showing islands of malignant squamous cells (H and E, ×100). (c) Focal areas of highly pleomorphic tumor cells. (H and E, ×400)



**Figure 3:** (a) Left ovarian peritoneal surface covered with abundant keratinous material (H and E, ×100). (b) Foreign body granulomatous reaction to the keratinous material (H and E, ×400)

## DISCUSSION

SCC is the most common malignancy arising from a teratoma followed by adenocarcinoma and melanoma.<sup>[2]</sup> It can rarely arise from squamous metaplasia occurring in the ovarian surface epithelium. Its treatment, outcome, and prognostic factors are not well understood, which pose great challenge and diagnostic dilemma.<sup>[2]</sup>

The clinical presentation of abdominal pain and distension is similar to other ovarian tumors, and bowel and bladder symptoms are seen in locally advanced cases. Patient age, tumor size, imaging characteristics, and serum tumor marker levels are the risk factors for malignancy in a teratoma.<sup>[1,2]</sup> Most of these cases present in the fourth to fifth decades.<sup>[3]</sup> However, few cases have also been reported in young patients around 20 years of age.<sup>[4]</sup> The youngest reported case was 21 years old.<sup>[5]</sup> Tumors with a diameter larger than 9.9 cm or tumors with rapid growth are associated with an increased risk for malignant transformation.<sup>[5]</sup>

Serum carcinoembryonic antigen and SCC antigen are the best screening markers than CA-125 and CA-19.9.<sup>[3]</sup> Measurement of these antigens would provide a good preoperative risk assessment and help in differentiating between a teratoma and SCC. Patients with both antigens normal had a 5-year survival rate of 100%, whereas, in those cases where both are raised, it was only 13.9%.<sup>[5]</sup> Important radiological features supporting malignant transformation include the presence of a solid component that extends transmurally invading the adjacent structures, necrosis, and hemorrhage.<sup>[6]</sup>

Grossly, SCC is a large cystic tumor with significant solid component. In one case series by Kido *et al.*, five of the six tumors had an obvious large solid component. Four of the five solid components exhibited transmural extension.<sup>[6]</sup> In our case, abundant keratin-filled pockets in the pelvis were mistaken for caseous necrosis, and a clinical diagnosis of tuberculosis was made. Focal tearing, localized rupture, and penetration of capsule by the tumor lead to the deposition of keratin debris and/or formation of keratinous nodule in the peritoneal cavity or adjacent organs. This can be mistaken for tuberculosis or metastatic malignancy of squamous differentiation.<sup>[7]</sup> Mostly, SCCs arising in teratomas are moderate to poorly differentiated.<sup>[2]</sup> In a study comprising 12 cases by Rekhi *et al.*, eight were moderately differentiated, three were poorly differentiated, and only single case of well-differentiated SCC was seen.<sup>[2]</sup> Poorly differentiated SCCs require p63 and CK 5/6 immunohistochemical stains for diagnosis.<sup>[4]</sup>

Tumors confined to the ovary are optimally managed with surgical management and close follow-up. There

may be rapid progression and dissemination of tumor after spillage of cyst contents at surgery or laparoscopy.<sup>[8]</sup> Adjuvant chemotherapy or radiotherapy is not beneficial in patients with Stage I or Stage II diseases, but it increases the survival in those with Stage III and Stage IV disease.<sup>[9]</sup> However, Rekhi *et al.* noted a good control of the disease with paclitaxel-based chemotherapy, as compared to the conventional chemotherapy regimen comprising cisplatin, etoposide, bleomycin, and vincristine.<sup>[2]</sup> The role of neoadjuvant chemotherapy is debatable. In a study by Abhilasha *et al.*, two of the ten patients were given neoadjuvant chemotherapy and showed partial to no response.<sup>[10]</sup>

## CONCLUSION

Preoperative diagnosis of SCC arising in the MT of the ovary is challenging. Despite thorough investigations, it may not be possible to differentiate between an ovarian malignancy and abdominal tuberculosis without exploratory laparotomy and histopathological examination. Therefore, an ovarian teratoma in a patient aged >40 years with huge size and/or extensive solid component should raise the suspicion of malignancy. Such specimens should be thoroughly sampled, and the patient must be followed up for a long period. More evidence for the treatment and management plans of malignant neoplasm arising from MT by multicenter studies is still 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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## Conflicts of interest

There are no conflicts of interest.

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