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CASE REPORT OPEN ACCESS

Early Recurrence in Case of Squamous Cell Carcinoma Arising from Teratoma Post-Surgery and Adjuvant Chemotherapy: Two Case Reports and Literature Review

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ABSTRACT

Background: Malignant transformation in mature cystic teratoma (MCT) is a rare phenomenon, with squamous cell carcinoma (SCC) being the most frequent histologic subtype. Its diagnosis remains challenging due to nonspecific clinical and radiological findings, and the prognosis is generally poor.
Aim and Objective: To study the early recurrence in case of squamous cell carcinoma arising from teratoma post-surgery and adjuvant chemotherapy: two case reports and literature review.
Case Summary: We present two cases—one ovarian and one extra-gonadal teratoma—with histologically confirmed SCC, both managed with surgery followed by adjuvant Paclitaxel-Carboplatin chemotherapy. Despite this standard management, both cases showed early locoregional recurrence within weeks of treatment completion, highlighting the aggressive course of such malignancies.
Conclusion: These reports underscore the importance of early suspicion, aggressive surgical intervention, and the need for a more tailored adjuvant treatment strategy in SCC arising from teratomas. Further studies are warranted to identify prognostic biomarkers and optimal post-operative management.

Keywords: Squamous cell carcinoma, mature cystic teratoma, malignant transformation, ovarian tumor, extra-gonadal teratoma, recurrence, chemotherapy.

INTRODUCTION

Mature cystic teratoma (MCT), commonly known as dermoid cyst, is the most frequent germ cell tumor of the ovary and accounts for 10–20% of all ovarian neoplasms. Most MCTs are benign and occur predominantly in women of reproductive age. However, malignant transformation occurs in 1–2% of cases, with squamous cell carcinoma (SCC) comprising over 80% of these rare events. The clinical presentation of malignant transformation within MCT is often insidious, with symptoms such as abdominal pain, distension, or a palpable mass mimicking benign tumors. Due to the lack of reliable preoperative markers or imaging features, diagnosis is frequently delayed until after surgical excision and histopathological analysis [1].

The pathogenesis of SCC in MCT remains poorly understood, but potential contributing factors include long-standing lesions, chronic irritation, and possibly human papillomavirus (HPV) infection. Prognostic indicators like patient age,

tumor size, tumor grade, and FIGO stage have been suggested in literature, but standardized management protocols are still lacking due to the rarity of the disease.

Teratoma with malignant transformation in gonadal or extra-gonadal sites is an uncommon presentation. Mediastinum is the most common site of Extra-gonadal teratoma with malignant transformation. Mature cystic teratoma of the ovary (MCTO) is a benign tumor that may develop in 10–20% of women during their lifetime, commonly in the 20-40 year age group [1]. Malignant transformation of MCTO was noted in only 1.4% [2]. Squamous cell carcinoma (SCC) constitutes 0.3% of malignant transformation, accounting for 80% of malignant transformation in MCTO [3,4]. SCC arising in MCTO is commonly observed in postmenopausal women [5]. The other histological types include adenocarcinoma, small cell carcinoma, sarcoma, malignant melanoma and mixed type [6]. The clinical presentation of SCC transformation in MCTO is not specific, making it difficult to diagnose preoperatively. The preoperative imaging studies and tumor markers are not specific to predict malignant transformation. The prognosis is usually poor, which depends on complete cytoreduction, age and stage of the disease. Adjuvant treatment with platinum-based chemotherapy has been shown to improve survival [3]. Here we present a two case report of squamous cell carcinoma arising from teratoma, one gonadal and one extra-gonadal, who were treated with surgery and adjuvant Chemotherapy.

We report two cases—one gonadal and one extra-gonadal—of SCC arising from MCT, both of which experienced early recurrence following standard surgical and chemotherapeutic management. These cases highlight the need for heightened clinical suspicion, thorough surgical intervention, and reconsideration of adjuvant therapy strategies in such rare malignancies.

MATERIAL AND METHODS

This study is based on a retrospective review of two rare cases of squamous cell carcinoma (SCC) arising from teratomas—one gonadal and one extra-gonadal—managed at our tertiary care oncology center. After obtaining informed consent from both patients, relevant clinical data, surgical notes, radiological findings, histopathological reports, and follow-up records were collected and analyzed.

Patient Selection

Two female patients, aged 32 and 45 years, who were diagnosed with mature cystic teratoma (MCT) with malignant transformation into SCC between January 2022 and December 2023, were included.
The inclusion criteria

1. Histopathologically confirmed SCC arising within mature cystic teratoma
2. Underwent complete surgical resection followed by adjuvant chemotherapy
3. Recurrence documented within 3 months of adjuvant treatment completion
4. Clinical and Radiological Evaluation

Detailed history and physical examination were carried out. Radiological imaging included contrast-enhanced computed tomography (CECT) of the abdomen and/or thorax, and magnetic resonance imaging (MRI) as appropriate. Serum tumor markers such as CA-125, AFP, β-HCG, LDH, and SCC antigen were measured preoperatively and during follow-up when applicable.

Surgical Management

Surgical resection was performed as per the standard protocol. One patient underwent an en-bloc resection of the adnexal mass with adherent abdominal structures followed by total abdominal hysterectomy and colostomy. The other patient underwent thoracoabdominal excision of a cystic mass with resection of involved diaphragm, lung, and liver tissue. Intraoperative findings and resected specimens were documented.

Histopathological Analysis

Gross and microscopic evaluation of surgical specimens was performed by experienced pathologists. Histological features such as squamous epithelial nests, keratin pearls, presence of hair shafts, and organ infiltration were noted. Immunohistochemistry was conducted as necessary.

Adjuvant Therapy and Follow-up

Postoperative adjuvant chemotherapy was administered using Paclitaxel and Carboplatin regimen (six cycles). Follow-up imaging was done using CECT to evaluate disease status after completion of chemotherapy. Recurrences were confirmed with imaging and biopsy.

Data Collection and Literature Review
Clinical data were compiled in a case report format. A narrative literature review was performed using databases like PubMed, Scopus, and Google Scholar for studies on SCC arising from teratomas. Keywords included: "squamous cell carcinoma," "mature cystic teratoma," "malignant transformation," "ovarian dermoid," and "extragonadalteratoma." Relevant peer-reviewed articles published in English up to July 2025 were included for comparative discussion.

CASE 1
A 32-year-old lady, P3L3, k/c/o T2DM, presented with pain in the abdomen and bloating for 6 months. She was evaluated with a CECT abdomen which showed an adnexal mass. An exploratory laparotomy was done and due to dense adhesions further procedure was abandoned. Omental biopsy was taken and peritoneal lavage was done. Biopsy showed invasive omental deposits by well differentiated squamous cell carcinoma. She was referred to our center for further management.

She was further evaluated with CECT Abdomen (Fig. 1) showing large irregular heterogeneously enhancing solid cystic lesion with multiple air foci and enhancing septations noted in left adnexa measuring 10.6x14x8.9 cm. Superiorly the lesion extends upto umbilical region and shows loss of fat planes with adjacent small bowel loops. Inferiorly the lesion shows loss of fat planes with the left lateral body of the uterus. Anteriorly it is opening into the subcutaneous plane of the anterior abdominal wall through a defect of length 1.6 cm.

CA 125 was 478.7 U/ml; Beta HCG was 0.6 IU/L and AFP was 5.4 ng/ml.
Abdominal examination revealed previous surgery scar mark with wound discharge and vague hypogastrium lump of size 10x10 cm.

Intraoperatively an enterocutaneous fistula noted at the previous Pfannenstiel incision. A 10x2 cm subcutaneous abscess cavity noted below the incision. Infraumbilical rectus sheath sloughed off and retracted. No ascites, peritoneal deposits. 15x15 cm soft to firm mass noted in left adnexa infiltrating and opening into infra-umbilical anterior abdominal wall with adherence to omentum, part of distal jejunum, proximal ileum and sigmoid colon. She was operated with En-bloc excision of left adnexal mass with adherent anterior abdominal wall, omentum, distal jejunum, proximal ileum and sigmoid colon and total abdominal hysterectomy with left Salpingo-oophorectomy with Jejuno-ileal, Ileo-ileal and colorectal anastomosis and diversion transverse colostomy.

Post-operative biopsy showed 12.7x11.5x7.5 cm tumor in the left ovary, well differentiated squamous cell carcinoma, arising from primary teratoma. Multiple sections from the tumor showed nests of malignant squamous cells with well-defined keratin pearls with infiltrative borders, infiltrating into the wall of adherent bowel. Grossly tufts of hair were identified within the cystic space.

She was staged as pT2bN0M0 and after MDTB discussion planned for adjuvant Paclitaxel and Carboplatin. She received 6 cycles of chemotherapy.
Within 2 weeks of completion of the last cycle, she presented with surgical wound site discharge and was evaluated with CECT Abdomen (Fig. 2) which showed intraabdominal and abdominal wall recurrence. She was planned for palliative chemotherapy.

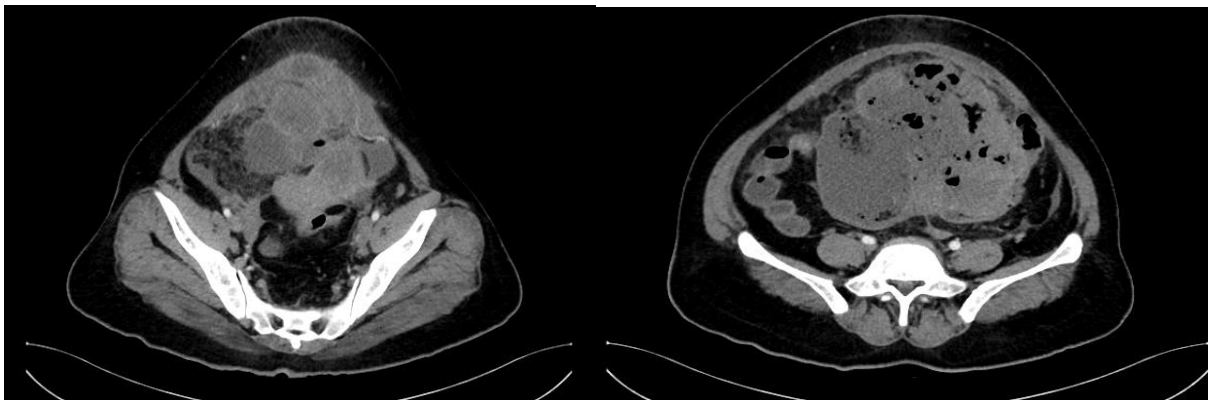

Fig. 1 Preop CT

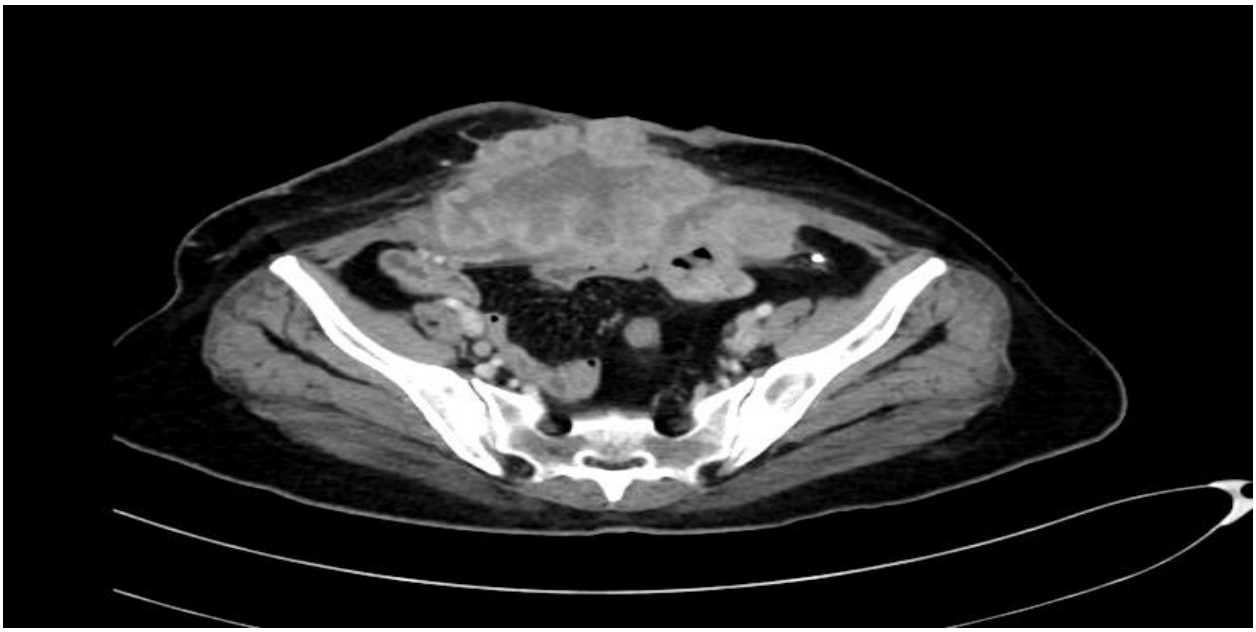

Fig.2 CT showing recurrence

CASE 2

A 45-year-old lady, known as a case of dermoid cyst of the left hemithorax operated in 1996, presented with radiating pain along the right upper limb.

CECT Thorax and Abdomen (Fig. 3) showed a 15.9x14.2x14.12 cm lesion in the right hemi thorax with two well defined heterogeneous area extending anteriorly-superiorly to 4th to 5th rib space with atelectasis of lungs parenchyma, medially abutting IVC, post laterally soft tissue and inferiorly abutting diagrammatic pleura.

MRI showed hetero intense enhancing lesion in right sub diaphragmatic region with solid component having liver infiltration of segment 7 – s/o malignant transformation.

ELISA (echinococcosis) was negative; Beta HCG was 14.0 IU/L; AFP was 1.4 ng/ml; LDH was 138 IU/L.
Biopsy showed multiple keratin flakes with keratin pearls.

Clinical examination of chest and abdomen did not reveal any significant findings.
She was operated with Excision of cyst by thoracoabdominal approach with mesh repair of diaphragm and Wedge resection of Right lung lower lobe and liver

Intraoperatively 20x15 cm mass occupying the lower thoracic cavity, adherent to the IVC and right hepatic vein medially, thoracic wall laterally and right lower lobe superiorly was noted. It was found infiltrating the diaphragm and segment 7 of the liver inferiorly.

Post-operative biopsy showed well differentiated squamous cell carcinoma arising in a background of mature cystic teratoma. Multiple sections showed predominantly keratin, areas of bronchial epithelium, hair shafts and gastric epithelium. Also seen are nests of malignant squamous epithelial cells with keratin pearl formation. Giant cell reactions to keratin and hair shafts are also noted.

Case was discussed in a MDTB and planned for adjuvant Paclitaxel and Carboplatin. She received 6 cycles of chemotherapy.

After completion of chemotherapy, follow up CECT (Fig. 4) showed suspicious liver deposit. CT guided biopsy was done which confirmed recurrence. She was planned for palliative chemotherapy.

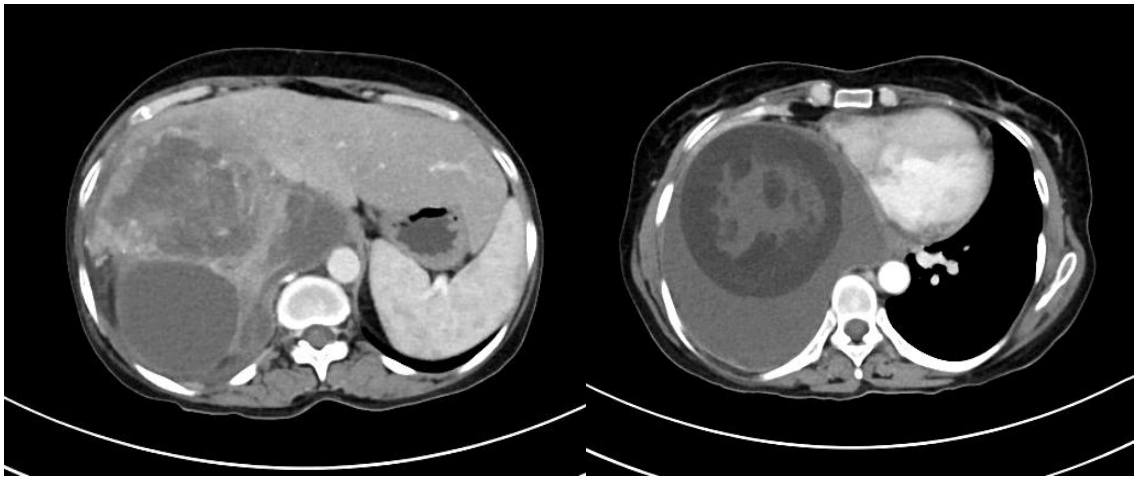

Fig. 3 Preop CT

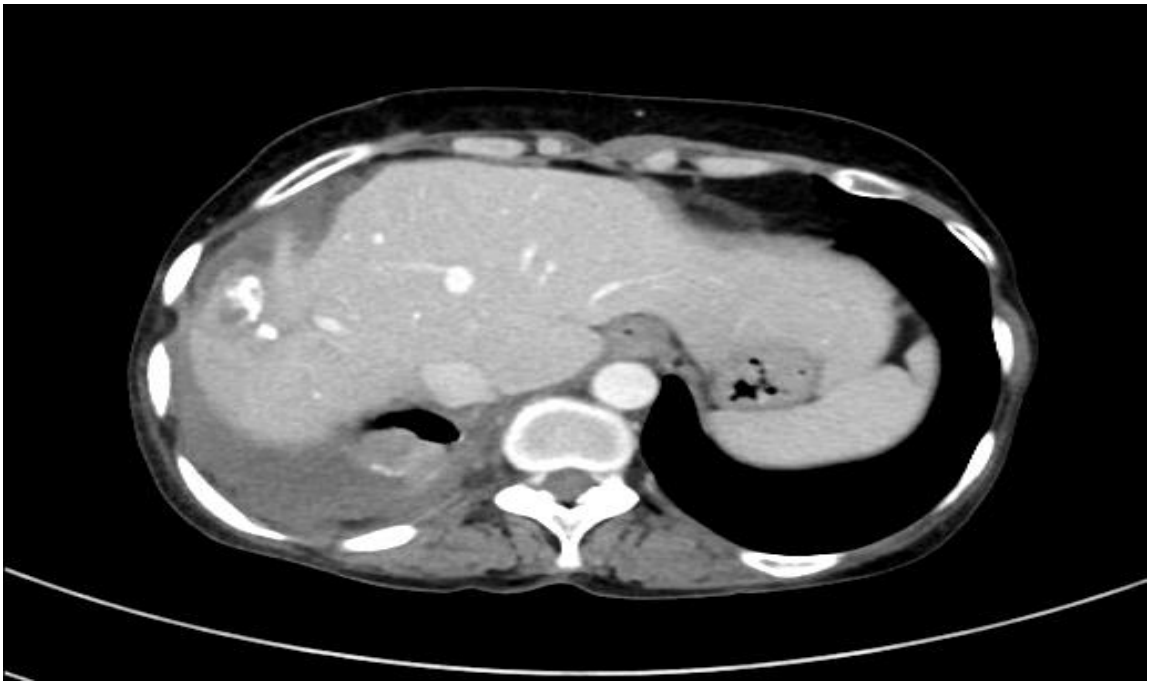

Fig. 4 CT showing recurrence

DISCUSSION

Squamous cell carcinoma is the most common type of malignant transformation arising in MCTO [4]. Other histological variants are thyroid carcinoma, carcinoid, papillary renal cell carcinoma, medulloblastoma and intestinal-type mucinous adenocarcinoma [7]. The etiology of SCC transformation in MCTO is not clear. SCC transformation in MCTO may be due to a continuous process of squamous metaplasia, atypical hyperplasia, carcinoma in situ, interstitial infiltration and invasive cancer [8]. A study in Taiwan has shown an association of SCC in MCTO with high-risk human papillomavirus (HPV) infection [9]. Malignant transformation is reported to occur in 1.4%, especially in an elderly woman with unilateral tumors and rapid growth [2,10]. Pure de-novo SCC of ovaries is uncommonly reported [11].

SCC arising from a MCT in most instances is not diagnosed preoperatively like our case. No specific clinical signs and symptoms designate a MCT to have undergone a malignant transformation. The literature reports these patients to have abdominal distension and bloating sensation along with the pelvic mass, similar to a benign MCT [12]. In a systematic review of SCC transformation in MCTO, 47.3% presented with abdominal pain and 26.0% with abdominal mass. Our 1st patient initially presented with abdominal pain and bloating for 6 months [3].

The role of tumor markers for diagnosis and surveillance of SCC transformation in MCTO is not well established. SCC antigen was found raised in 16 out of 24 cases of SCC arising in MCTO in a study in Taiwan. SCC antigen was found elevated during recurrence in the serial monitoring [13]. In a systematic review, median preoperative SCC antigen was 7.4 ng/mL, CA 125 was 64.4 U/mL, CA 19-9 was 144.0 U/mL and CEA was 6.9 ng/mL [3]. In our first patient, CA 125 was raised. Beta HCG and AFP were normal in both patients.

Doppler imaging studies revealing vascularization can be a useful indicator of malignant transformation [14,15]. CT showing adnexal mass with fat/fluid level and fat droplets would suggest dermoid cyst [16]. It is very difficult to detect malignant transformation of MCT in preoperative radiological scans. CT scan and magnetic resonance imaging (MRI) have no role in detecting malignant transformation. In our cases, Doppler study was not done as both the masses were not suspected to have malignant transformation. Hence there is a need to have objective ways to lay down the risk assessment of MCT having chances of a malignancy, as it is a common tumor.

The conventional prognostic markers of malignant MCT include tumor size, old age, solid components, FIGO stage, intraperitoneal rupture of cyst, grade, lymphovascular invasion and pattern of tumor infiltration (infiltrating/broad front) [17]. Peterson et al in a study of 190 malignant teratoma have reported a very high metastatic rate (64%) [18]. In a systematic review, tumor size ⩽10 cm and >10 cm did not show any survival differences [3]. Study by Kikkawa et al reported that the chances of MCT are higher when a patient's age is higher than 45 yrs and tumors larger than 9.9 cm are more prone to develop malignancies with a sensitivity of 86% [19].

Surgery forms the mainstay of management of teratomas with malignancies [17,18]. Although comprehensive staging laparotomy is standard care for ovarian cancers, a systematic review has found that hysterectomy and omentectomy have shown survival benefit, but lymphadenectomy was not shown to improve survival in cases of SCC transformation in MCTO [3]. In another systematic review, omentectomy did not show survival benefits, whereas lymphadenectomy improved chances of survival in advanced cancers [20]. In nulliparous women especially in the reproductive age group having early stage malignant teratoma, a simple conservative unilateral oophorectomy can be offered. However, in the postmenopausal women, radical surgery like total abdominal hysterectomy with bilateral salpingo-oophorectomy is planned [17]. Post Surgery combination chemotherapy, radiotherapy, or both are used. No large trials have been carried out in these sets of patients to have a consensus on chemotherapy/ radiotherapy protocols [18]. Our first patient underwent En-bloc excision of left adnexal mass with adherent anterior abdominal wall, omentum, distal jejunum, proximal ileum and sigmoid colon and total abdominal hysterectomy with left Salpingo-oophorectomy and second patient underwent Excision of cyst by thoracoabdominal approach. Both of them were given 6 cycles of Carboplatin and Paclitaxel as adjuvant. Patients with malignancies in benign teratoma have a very grave prognosis with a 5-year survival of 20%.

Malignant transformation in MCT is a rare but lethal event. SCC is the most prevalent histological variant reported in over 80% of such cases. Our findings align with the literature, where patients often present with vague symptoms such as abdominal pain or fullness, making preoperative diagnosis difficult. Advanced imaging modalities and tumor markers have limited utility in detecting malignant transformation before surgery.

Recent studies have examined the association between HPV and SCC in MCT. Zhang et al. (2024) found a significant correlation between high-risk HPV subtypes and malignant transformation in ovarian teratomas, suggesting viral oncogenesis as a possible mechanism [21].

In terms of imaging, modern radiomics-based approaches have attempted to differentiate benign from malignant teratomas. A study by Patel et al. (2025) [22] demonstrated promising results using artificial intelligence-enhanced CT scans to detect malignancy in complex ovarian masses, though further validation is required.

The role of tumor markers such as SCC antigen remains controversial. Lee et al. (2024) [23] reported elevated SCC antigen in 70% of MCT-associated SCC cases and suggested its use in both diagnosis and monitoring for recurrence.

Surgical resection remains the cornerstone of management. As seen in our cases, extensive surgical debulking was performed, and histology confirmed well-differentiated SCC arising from MCT. Adjuvant chemotherapy with Paclitaxel and Carboplatin, while standard, did not prevent early recurrence in either case, emphasizing the aggressiveness of the disease.

Recent studies indicate that the benefit of adjuvant chemotherapy may vary depending on tumor stage and grade. Yamamoto et al. (2025) [24] found that SCC arising in MCT with stage I-II disease might benefit from surgery alone, whereas stage III-IV tumors required combination chemotherapy for improved survival.

Additionally, immunohistochemical markers such as p16 and Ki-67 have been studied for their prognostic utility. A multicenter study by Singh et al. (2024) [25] reported that high Ki-67 index correlated with early recurrence and poor overall survival.

Despite treatment, the prognosis remains grim. Our patients experienced recurrence within 4 weeks of completing chemotherapy. This is consistent with global data showing a 5-year survival rate of only 20–30% for SCC-transformed teratomas. Furthermore, recurrence patterns are often locoregional, as in our cases, indicating the aggressive local behavior of these tumors.

Emerging therapies such as immune checkpoint inhibitors and targeted therapies are under investigation. A phase II trial by Costa et al. (2025) [26] is evaluating the role of pembrolizumab in recurrent SCC-MCT, with early data showing disease stabilization in select patients.

Both of the index patients were treated with surgery f/b adjuvant chemotherapy and both of them developed recurrence within a month of treatment completion. The patient with gonadal MCT developed intraabdominal and abdominal wall recurrence. Second patient with extragonadal GCT developed liver metastasis. Both of these were locoregional recurrences implying the locally aggressive nature of the tumor.

CONCLUSION

It is challenging to come to a definite preoperative diagnosis of malignant transformation in MCTO. Therefore, clinicians should be aware of this rare entity and should have high suspicion of malignant transformation when a postmenopausal woman presents with a large tumor size with irregularly thickened cystic wall or solid foci on the tumor wall. The gynecologist/oncologist and pathologists should always entertain a differential of a malignancy in a teratoma when the patient is old, has an unusually large cyst and has raised serum tumor markers. Surgery f/b adjuvant chemotherapy is the preferred treatment modality, though there are no large clinical trials showing benefit of adjuvant therapies. Prognosis of MCT with malignant transformation is poor with a 5-year survival of 20%.

LIMITATIONS

1. This report includes only two cases, limiting generalizability.
2. Lack of molecular and genomic profiling restricts insight into tumor biology.
3. Short follow-up period prevented long-term outcome analysis.
4. Immunohistochemistry for p16, Ki-67, or HPV was not performed due to resource constraints.
5. Absence of standardized treatment guidelines for SCC in MCT hinders therapeutic comparison.

DECLARATIONS:

Conflicts of interest: There is no any conflict of interest associated with this study

Consent to participate: There is consent to participate.

Consent for publication: There is consent for the publication of this paper.

Authors' contributions: Author equally contributed the work.

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