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Suqamous Cell Carcinoma Of The Ovary Arising From Mature Cystic Teratoma With Metastasis To The Mediastinum

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Abstract: *Malignant transformation in a mature cystic teatoma (MCT) of the ovary is a rare complication, occurring in only 1–2% of cases, with squamous cell carcinoma (SSC) being the most common type. Preoperative diagnosis is difficult because of the absence of specific symptoms and signs predicting malignancy. A 78-year-old women presented with lower abdomen mass. Ultrasound and CT scan revealed complex pelvic mass measuring 15 cm. We performed complete cytoreduction, including an en-bloc resection of the tumor and rectosigmoid colon, omentectomy and pelvic-paraaortic lymph node dissection. Histopathology revealed SSC arising from a MCT of the left ovary penetrating the rectal wall. she received 2 cycles of adjuvant platinum-based chemotherapy. She developed 2 months later mediastinum metastasis and had chest irradition. Size of the tumor and patient's age should be regarded as important predictors of malignant transformation in MCT. Histopathological examination has an important role in the diagnosis and prognosis of this rare tumor.*

Keywords*: mature cystic teratoma, malignant transformation, squamous cell carcinoma.*

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I. Introduction

Squamous cell carcinoma (SCC) of the ovary can arise from mature cystic teratoma, ovarian endometriosis, a Brenner tumor, or solely from an ovarian surface epithelium [1]. Mature cystic teratoma (MCT) is composed of well-differentiated tissues derived from the three germ cell layers (ectoderm, mesoderm, and endoderm)[2]. Malignant transformation of the mature elements within the MCT is an extremely rare complication, occurring in only 1–5% of all cases [3-5]. Squamous cell carcinoma accounts for 70– 88% of all malignant tumors arising in the mature cystic teratomas followed by other rare malignancies including adenocarcinoma, carcinoma, malignant melanoma, transitional cell carcinoma, thyroid adenocarcinoma and sarcomatous changesthe much rarer adenocarcinomas and carcinoids [1, 5].

Because of its rarity, clinical presentations, preoperative risk assesement, treatment and prognosis are poorly understood.[1] Therefore, we report a case of a woman with SSC arising in MCT with focus study on the value of preoperative prediction of this malignant transformation, treatement approach and prognosis factors.

II. Case Report

A 78-year-old tunisian woman (gravida,4 ; para, 3) presented with abdominal mass that had been discovered several weeks earlier. The patient went through menopause at the age of 50 years, and had no previous exposure to any hormonal treatment. She had a past medical history of hypertension and type 2 diabetes and had undergone a cholecystectomy and an appendicetomy, 2 years previously. There was no family history of gynecological malignancy. Physical examination revealed a large ferm, mobile pelvic mass mesuring 20 cm. Ultrasonography detected a 15 cm complex mass with solid components probably ovarian. the endometrium was thin and clear. The patient's serum CA-125 concentration was 22.2 U/mL. CT scan showed a suspicious 15 cm mass of the left ovary with cystic and solid components (Fig 1).

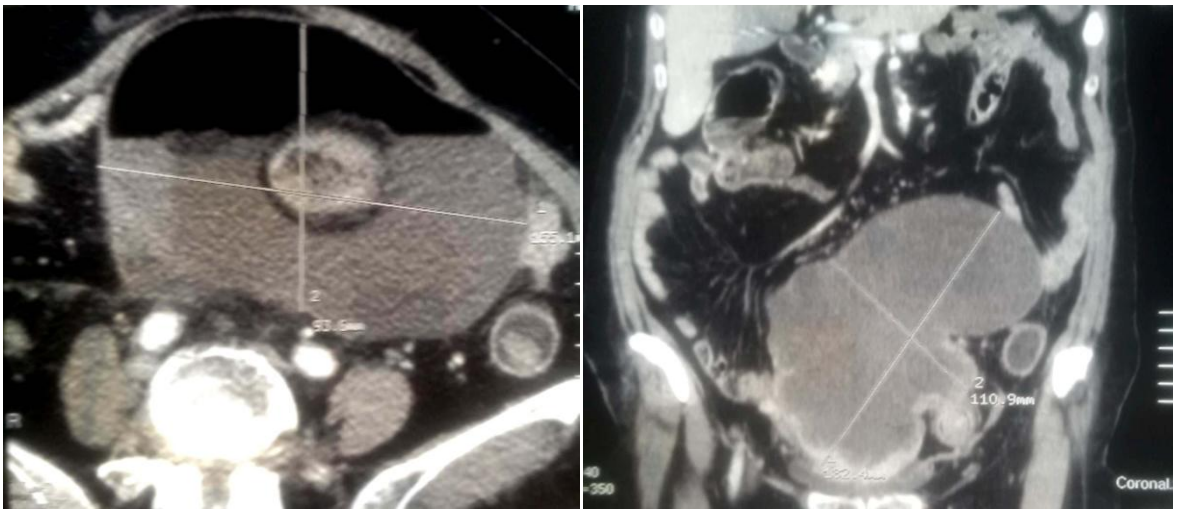

Fig1 : a 15 cm mass of the left ovary with cystic and solid components.

An exploratory laparotomy was performed. There was no ascite and no abnormal peritoneal lesions in the pelvis or abdomen. The left ovary was replaced by a tumour measuring 15 cm which was fixed to the rectal sidewall. A unique metastatic implants of 1 cm was found on the ilieum 25 cm from caecal bottom. There were no palpable pelvic nor paraaortic lymph nodes. The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy including an en-bloc resection of the rectosigmoid colon, omentectomy and pelvic-paraaortic lymph node dissection, a partial resection of the ileum resection, and infragastric omentectomy and pelvic and paraortal lym node dissection. there was no macroscopic residual tumor left following surgery. Her postoperative course was uneventful.

Postoperative pathology showed a well-differentiated, invasive SCCa arising in mature cystic tertoma of the ovary. there were no evidence of Brenner tumor or endometriosis. The cut surface of the ovarian mass was grayish white and showed hemorrhagic areas and patch necrosis. The ovarian capsule was involved in the mass. The fallopian tube was unremarkable. The tumor was composed of polygonal squamous cells that had individual cell keratinization and intercellular bridges, but keratin pearls were absent. The cellularity was high with nuclear pleomorphism and the mitotic figures were numerous. To exclude any other associated cervical lesions, her cervix and vaginal cuff were examined but there was no sign of tumor or dysplasia.Three pelvic lymphnodes were positive.

After classification of her tumor as an ovarian SCCa of International Federation of Gynecology and Obstetrics (FIGO) stage III, adjuvant chemotherapy consisting of six cycles of paclitaxel and carboplatin was indicated but she received only 2 cycles. She developed two months later chest pain. CT scan revealed a solid mediastinal metastasis mesuring 3cm (Fig 2). Radiotherapy to the mediastinum was administered in 10 fractions of 2 Gy. She was last seen one month after radiotherapy when she refused any further treatement.

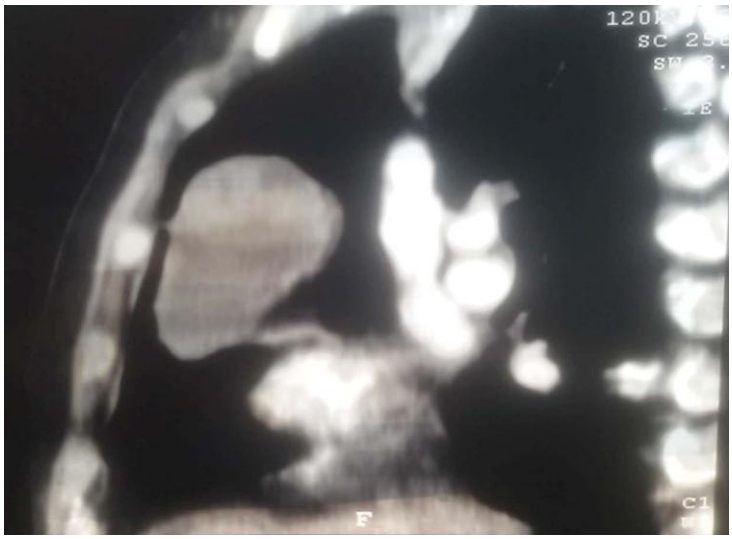

Fig 2: solid mediastinal metastasis

III. Discussion

Mature cystic teratomas are part of a subgroup of ovarian germ-cell tumour believed to arise from the primordial germ cells. Ovarian germ-cell tumours represent 20–25% of ovarian neoplasms and 5% of ovarian cancers[6]

Mature cystic teratomas are very common in women of childbearing age and they are bilateral in 10–17% of cases[1, 6]. Most mature cystic teratomas are detected 15–20 years before they undergo secondary malignant transformation because cytogenetic abnormalities precede many years histological changes[5, 6]. Malignant transformation of MCT typically occurs in women between 40 and 60 years of age [3]. Thus, squamous-cell carcinoma arising in mature cystic teratoma is more common in postmenopausal patients [1, 6, 7].

Histogenetically, two hypotheses might explain the histogenesis of SCC in MCT. They has been considered to arise either from the epidermis or from the respiratory epithelium [3]. Furthermore, high-risk human papillomavirus infection might be a causal agent associated with malignant transformation[6, 8].

The clinical features are non specific and similar to other ovarian tumors. they usually include abdominal pain and distension secondary to a pelvic mass, but it may also present with bowel or bladder symptoms in cases of locally advanced disease [4, 7]. Cyst rupture or ovarian torsion can be the revealing symptom for large tumors [8, 9].

Since MCT is a common ovarian neoplasm and is frequently diagnosed in young women, there is a growing interest in preoperative risk assessment of malignancy of these tumors in order to optimize treatement. Risk factors for malignancy in MCT include patient's age, tumor's size, imaging characteristics, and serum tumor markers [4].

Takashina et al. demonstrated the role of patient's age in differential diagnosis, with a mean age at diagnosis of 32 years for MCT versus 50 years for SCC. They noted that a higher suspicion of malignancy in MCTs was occurring in patients over the age of 45[10].

Tumor size has also been noted to predict malignancy. In Rathore's study, the average size of malignant tumor was 11.7 cm, compared to 7.6 cm in MCT [5]. Kikkawa et al, reported that a tumor diameter of > 9.9 cm was 86% sensitive for malignancy in their series [2]. These results may suggest that MCT which has undergone malignant transformation will typically be larger than a benign MCT and should be treated as early as possible [1].

Among four tumor's markers (SCC antigen, CA125, CA199, and CEA) used in diagnosis of squamous cell carcinoma, SCC antigen had the highest positive rate [1, 2, 8]. Mori et al, noted that the use of a single factor is not particularly effective. they found that the combination of SCC antigen level inferior to 2.5 ng/ml and age less of 40 years has a 77% sensitivity and 96 % specifity in predicting malignant transformation[11].

Modern imaging techniques, such as MRI, may be more useful in preoperative risk assessment of malignant transformation[9, 12]. Neovascularization in tumors has been histologically shown to consist of primitive thin-walled vessels with a paucity of smooth muscle[4]. Kido et al, studied the MRI appearance of SCC arising in MCT and correlated malignancy with the presence of a solid component with contrast enhancement, transmural extension, evidence of adherence to surrounding structures, hemorrhage, and necrosis [13].

However all these preoperative diagnostic procedures are not certain in ruling out malignant transformation, and ovarian tumors should be considred potentially malignant until histologically proven benign, despite the patient's age, tumor's size, imaging inverstigations and tumor markers profiles [6].

Making a differential diagnosis remains very important because surgical management is quite different for MCT and SSC ,especially, laparoscopiy which is frequently performed in case of MCT, but can be risky when malignant transformation is suspected [2].

There is no clear consensus on optimal management strategy for these patients. Most authors would advocate that surgery should be the initial treatment [7]. Surgical treatment of SSC arising in a MCT is similar to that of epithelial ovarian cancer according to litterature. Young Patients with stage IA disease and for fertility considerations, conservative treatment with unilateral salpingo-oophorectomy, surgical staging, and close follow-up has been proposed [4, 6]. The benefit of lymph node dissection is controversial since the SSC spreads transmurally with extensive local invasion or peritoneal seeding, but it may influence treatment planning, especially for early-stage disease[4, 5] Moreover, Hachkethal et al, suggested that lymphadenectomy improved overall survival in patients with stage II and above disease, in patients (59.2 months of mean OS in lymphadenectomy group vs 40.4 months of mean OS in no lymphadenectomy group), although only 14% of these cases underwent an adequate staging surgery [6]. For advanced-stage disease , optimal debulking was associated with increased survival[9]. Optimal debulking if metastatic disease is present is also thought to improve survival , as it does for primary epithelial tumours[7].Our patient underwent The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy including an en-bloc resection of the

rectosigmoid colon, omentectomy and pelvic-paraaortic lymph node dissection, a partial resection of the ileum resection, and infragastric omentectomy and pelvic and paraortal lym node dissection.

Adjuvant therapy for SSC has not been defined. The current recommendation of cisplatin is related to its activity in ovarian cancer and gynaecological squamous-cell carcinomas[2, 9]. Although SSC are originally sensitive neoplasmS for irradiation, the benefit of pelvic radiotherapy remains unclear[10]. Concurrent chemoradiation has also been advocated by several authors in a similar manner to that used for squamous cell carcinomas of the cervix disease [4, 7]. In a case series of 17 patients Do santos et al, studied retrospectively the impact of adjuvant chemotherapy combined or not with radiotherapy on survival. Six out of 17 patients received adjuvant chemotherapy and 4 chemoradiotherapy. Although no conclusions were made since the median survival was not reached, they recommanded the use of platinum-based chemoradiotherapy combined with external pelvic radiation for early-stage disease [4]. Alkylating drugs should be considered for chemotherapy regimens[6].

SSC has generally poor prognosis when disease has spread beyond the ovary [4]. The prognosis depends mainly of tumor's stage[7]. Chen et al, analyzed 220 cases reported over a period of 30 years (from 1976 through to 2005).The 5-year survival rate was 100% for carcinoma in situ, 75.7% for stage I, 33.8% for stage II, 20.6% for stage III and 0% for stage IV. Elderly patients and large tumors were also two factors associated with poor prognosis. Preoperative tumor markers, SCC antigen and CA125, were also evaluated : the 5-year survival rate was 13.9% when they were both positive. This rate increases to 78.8%, if one of the two markers was positive, and to 100% when both were negative [1].

Hence, Age, tumor size, tumor markers level, extent of cytoreduction, FIGO stage and histologic caracteristics of the tumor (grade, capsular invasion, and the presence of vascular invasion) can provide valuable information for the prediction of patients survival in case of SCC arising from MCT [2, 4, 8].

IV. Conclusion

Malignant transformation occurs in 1-2% of MCT s of the ovary and usually consists of SSC. Because of the rarity of this malignant transformation, The preoperative diagnostic is quite difficult. There is no consensus regarding optimal management. thus, diagnosis and treatment remain a challenge for gynecologic oncologists.More studies and clinical trials are needed to establish the best treatment options for patients with this disease. In the meantime we recommend vigilant preoperative risk assessment, surgical cytoreduction with proper staging, and adjuvant therapy with platinum-based chemotherapy. The role of concurrent whole pelvic radiation, in surgically confirmed early stage disease, remains unclear.

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