

**CASE REPORT**

Squamous Cell Carcinoma Arising From Mature Cystic Teratoma of Ovary

Vijaylaxmi V Suranagi, Prakash R Malur*

Abstract

Malignancy occurring in a mature cystic teratoma is extremely a rare entity. Preoperative diagnosis of malignant transformation with in a mature cystic teratoma is extremely difficult and poses a great challenge to current clinical surgical practice. Factors such as age, tumor size, tumor markers and tumor imaging characteristics help in preoperative risk assessment. It is important to be aware of this entity when cystic teratomas are encountered in a peri or postmenopausal women for proper management of the case. We encountered a 45 years female, with vaginal bleeding. Right ovary was enlarged. The mass excised from the right ovary had cystic and solid areas. Its wall was markedly thickened. Histopathological examination of the cyst revealed an invasive squamous cell carcinoma in a preexisting mature cystic teratoma. A thorough search for primary focus of squamous cell carcinoma elsewhere was made to rule out a possibility of metastasis into the ovary.

Key Words

Squamous Cell Carcinoma, Mature Cystic Teratoma, Ovary

Introduction

Cystic teratoma, a tumor arising in young women are usually benign, but rarely may be associated with germ cell tumor or exhibit secondary malignancy after initial growth. Primary squamous cell carcinoma (SCC) of the ovary is extremely rare (1). Few cases reported have been malignant arising from either a cystic teratoma or a Brenner tumor, without a cervical malignancy. Malignant transformation occurring in mature cystic teratoma (MCT) is again rare and found to be 1-2% of all mature cystic teratomas. Prognosis of patients with squamous cell carcinoma of the ovary are quite poor. Preoperative assessment of the risk of malignancy is extremely difficult but very important for proper treatment planning and management. We report one such rare case of primary ovarian squamous cell carcinoma arising in a cystic teratoma, encountered in a peri menopausal woman which seems to be worthy of review. Also a brief review of literature with emphasis on factors which help in preoperative risk assessment is presented.

Case Report

A 45 years female (P3L3) presented with history of pain in the right lower abdomen with per vaginal bleeding

and back ache for 2 months. On examination, tenderness was noted on the right side of the lower abdomen. Abdomen was tense, no mass was palpable. Per vaginal examination revealed fullness of the posterior fornix where a nodular mass was felt. Preoperative clinical diagnosis of infected dermoid cyst was rendered. Hysterectomy with bilateral salpingo-oophorectomy was performed. Post operative period was totally uneventful.

The uterus, cervix and left ovary were normal. Right ovary was enlarged, measured 10x5x3.5cms. External surface was irregular and covered with inflammatory exudate. Cut surface of mass revealed it to be cystic with prominent solid areas. The cyst wall was markedly thickened, irregular. The cavity contained pultaceous material, hair and tooth (*Fig. 1*) Microscopic examination revealed a malignant neoplasm with features of squamous cell carcinoma, invading the wall of the cyst, displaying various stages of development from dysplasia, carcinoma in situ to frankly invasive areas. The cyst wall showed invasion by the neoplastic cells, up to 2/3rd of its thickness. However there was no breach in the serosal surface (*Fig. 2*).

From the Deptt. of Pathology and *Urology JN Medical College and KLE Prabhakar Kore Hospital &MRC, Belgaum-Karnataka
Correspondence to : Dr Vijaylaxmi V Suranagi A 14/11, JNMC Staff Quarters, Nehrunagar-Belgaum- Karnataka-India



A thorough search for evidence of malignancy in cervix was made with no effect. Careful head, neck examination, sinus and chest X- rays, pelvic examination did not show evidence of any non ovarian source of malignancy. She had no symptoms referable to her esophagus, pancreas or bladder. Finally, a diagnosis of primary squamous cell carcinoma arising from a cystic teratoma was given. The patient received radiotherapy following the diagnosis.



Fig.1. Cut Section of Cystic Ovarian Neoplasm Containing Necrotic Debris & Hair. Cyst Wall Show Marked Thickening

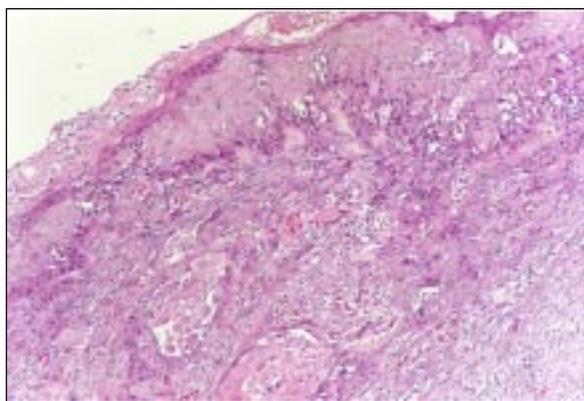


Fig.2 Cyst Wall lined by Squamous Epithelium Showing Varying Degree of Dysplasia & Invasive Keratinizing Squamous Cell Carcinoma. (H & E; 400X)

Discussion

Squamous cell carcinoma arising from MCT is extremely rare and is the most serious complication of this relatively common neoplastic lesion. Although MCTs are frequently bilateral, malignant change has usually been reported on only one side. SCC arising in MCT most probably develops from epidermal elements (80%) (2), although an origin from bronchial epithelium is a possibility.

In favour of former origin is finding of SCC in situ adjacent to carcinoma (3). Alternatively, SCC can arise from endometriosis or Brenner tumor. SCC may also be seen as metastatic deposits from SCC elsewhere especially cervix, even though that tumor may have occurred many years previously (4). Our patient however did not reveal such source. The carcinoma begin at or near dermoid protuberance, continue to grow without clinical evidence, eventually penetrating full thickness of the wall, developing direct extension and malignant adhesions to the neighboring organs (5). In 2/3rd cases, invasion or metastasis have occurred before diagnosis (2). Spread beyond the capsule can produce peritoneal seeding and symptoms such as pain, ascitis and signs of peritoneal irritation, such cases being prognostically poor. Several authors stress prognostic importance of intact capsule stating good prognosis if confined to the cyst (6). The diagnosis is frequently made unexpectedly in operating room or after final pathological examination, as in the present case. Preoperative diagnosis of malignant transformation within a MCT is extremely difficult, poses a great challenge and dilemma regarding a need for surgical staging and adjuvant therapy (1).

Risk factors for malignancy in MCT include patient age, tumor size, imaging characteristics and serum tumor markers. It has been observed that compared to benign MCT malignant transformation occurs in relatively older patient population, the mean age range reported being 45-60 yrs. Frequency of malignant change, increase with increasing age, rising to 19% in women after menopause (1,7). Hence, the need for the thorough search for malignant change, in dermoid cyst after the age of 45 years.

Larger tumors correlate with increased risk of malignant transformation. Kikkawa *et al* (8), in their case series observed that tumor diameter >9.9cm was 86% sensitive for malignant change. Importance of tumor markers is studied in many studies. According to Kikkawa *et al* (8) it was found that CEA was the best screening marker followed by SCC Antigen, both of these being superior to CA-125 and CA-19-9. It was finally recommended that measurement of CEA and SCC Ag levels in patients aged 45years or older, who have MCT like ovarian tumor larger



than 9.9cm in its greater diameter would provide a good clinical strategy for preoperative risk assessment and help in making differential diagnosis between MCT and SCC in MCT. In yet, another study Mori *et al* (9) reported that age >40years and serum SCC Ag >2.5ng/ml were 77% sensitive and 96% specific for malignant transformation which has also been useful in monitoring for recurrent disease. Tumor imaging characteristics which may aid preoperative risk assessment have also been studied. According to Lai *et al* (10) characteristic Computerized tomography findings are, adnexal mass with fat, calcification, soft tissue component and areas of invasion through the teratoma wall must raise a suspicion of associated malignancy. Kido *et al* (11) on Magnetic resonance imaging correlated malignancy with presence of solid component with contrast enhancement, transmural or trans-septal extension, evidence of adherence to the surrounding structures, necrosis and hemorrhage.

Santos *et al* (1) reviewed 7 case series with ≥ 10 patients with SCC arising in MCT. But because of relative rarity of these tumors, found that there was no uniform consensus regarding optimal management of such cases and also adjuvant therapy has not been preoperatively evaluated. It was suggested that when there is a preoperative suspicion of malignant transformation, optimal management include careful gross inspection of the tumor and frozen section analysis. An open approach is advisable to prevent upstaging by intra-peritoneal rupture during removal. Complete tumor excision and proper staging are integral to prognosis. Whole pelvis radiation and concurrent weekly platinum based chemotherapy may be beneficial in patients with stage I-II disease. Patient with stage III disease have poor prognosis and should be offered platinum based chemotherapy regimens.

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