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Carcinomatous Trasnformation In Mature Cystic Teratima of Bilateral Ovaries in Elderly Female

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ABSTRACT

Squamous cell Carcinoma of ovary is a rare occurrence and usually arises in mature cystic teratoma (MCT) or dermoid cyst of the ovary. The reported incidence of malignant transformation in a mature teratoma is approximately 2%. Though squamous cell carcinomatous change is the most common out of them, is still rare to find in bilateral ovarian dermoid cysts. A case of squamous cell carcinoma arising in dermoid cysts of bilateral ovaries presenting at an early stage is presented here. A 64 year old post menopausal lady, presented with complain of pain and discomfort lower abdomen since one and half month and a large complex abdominopelvic mass on examination and investigations. Final histopathology was reported as squamous cell carcinoma (SCC) arising from dermoid cyst of both ovaries. The patient was finally diagnosed as SCC of ovary arising in a MCT, assigned to surgical stage 1c2. In view of poor prognosis adjuvant chemotherapy was started.

KEYWORDS Mature cystic teratoma, ovary, squamous cell carcinoma, tumor markers.

Introduction-

Mature cystic teratomas (MCT, also called dermoid cysts) account for about 25% of all ovarian neoplasms and around 30% of all benign tumors arising in the ovary [(1)] with 0.8% - 5% (average of 1.4%) reported incidence of malignant transformation [(2,3)]

Squamous cell carcinoma (SCC) in MCTs is seen in 10-28% of post menopausal women. [(4)] There are no definite clinical features in these cases. Tumor markers are not often raised and imaging methods are many times not helpful. Hence, most cases are diagnosed postoperatively by histopathology. Tumors confined to the ovary usually have a better prognosis and patients with stage III/IV disease rarely survive five years. Very few cases have bilateral involvement. Here, we are presenting a case of SCC in dermoid cysts of both ovaries which was diagnosed at an early stage and was managed appropriately keeping in mind the poor prognosis of the condition.

Case Report-

A 64 year old postmenopausal lady, gravid 3 para 3 presented with complaints of pain and fullness in lower abdomen since one and half month. Her general condition was average. There was no visible or palpable evidence of any neck swelling, breast abnormality, or lymphadenopathy. Per abdominal examination showed a diffuse firm to hard, non-tender mass in bilateral lilac and hypogastric regions arising out of pelvis. Routine and specific investigations were carried out. Ultrasonography of whole abdomen suggested the possibility of dermoid cysts of both ovaries. MRI of abdomen reported a large complex multicystic abdomino-pelvic mass with fat fluid levels. Tumor markers were carried out and values were reported as follows: CA 125 : 1430 / ml, CEA :13.1 ng / ml, CA 19.9 : 2350 u / ml.

She was subjected to laparatomy. A large abdomino-pelvic, irregular, twisted and ruptured left ovarian mass (Fig.1b) and intact solid cystic right ovarian mass (Fig.1a)were found. The left ovarian mass was densely adherent to omentum, intestine, pelvic peritoneum and appendix. Adhesions were dissected and both the masses along with part of omentum were sent for histopathological study. Complete surgical staging and primary cytoreduction was done. Grossly right ovarian mass was intact, cystic measuring 6x4x2cm. On cross section wall was found to be thin and the cyst was filled with yellowish poltaceous material and hair follicles. One area was thickened. Right ruptured ovarian mass was large of 12x6x4 cm with solid and cystic areas and necrosis at places. Multiple sections were taken from both ovarian masses including the thick and necrotic areas. Histopathological sections revealed mature glandular elements with nests of malignant squamous cells with parakeratotic pearls. (Fig.2a &b) Final histopathological diagnosis was rendered as SCC arising from MCT of B/L ovaries. Both fallopian tubes, paramatrial tissues, peritoneal biopsy, uterus, omentum and pelvic lymphnodes were unremarkable without any evidence of malignancy. Peritoneal fluid was negative for malignant cells.

For management, the patient was assigned to SCC of ovary arising in a MCT, surgical stage 1c2. Her postoperative period was uneventful. In view of poor prognosis, adjuvant chemotherapy of six cycles of padi and carboplatin was started.

Discussion-

SCC of ovary is quite rare and usually arises in MCT of ovary (upto 2%). [(4)] Preoperative diagnosis is difficult due to poor screening methods MRI may be helpful. CEA may be elevated. Takagi et al have found CEA to be more useful than CA 125 and CA 19.9 in malignant transformation of MCT. [(5)] Other rare pathologies in MCT are adenocarcinoma and melanoma. Kar A et al had reported a rare case of carcinosarcomatous transformation in a mature teratoma of ovary comprising of squamous cell carcinoma with pleomorphic sarcomatous component showing myogenic differentiation[6]. The carcinomatous area was positive for high molecular weight keratin (HWK) and sarcomatous component was diffusely positive for vimentin. In the present case the tumor cells were positive for Cytokeratin. This entity carries poor prognosis specially when diagnosed in advanced stage, optimal cytoreduction is difficult to achieve in advanced disease. Suspicious features of malignant transformatin in dermoid cysts (clinical and on MRI) include: Postmenopausal women (mean age 55 years Vs 37.5

years for benign MCTs), large size of tumor (mean 15.2 mm Vs 88 mm for benign). solid areas within the cyst and invasion of adjacent organs / capsule[7,8]. Later in 2009 it was confirmed by Al Rayyan ES et al[9].

A systematic review and analysis of published data was done by Hackethal et al. (10) Sixty four suitable studies provided information on 277 patients. It was observed that SCC in MCT was mainly found in women above 50 years of age, having a high concentration of CA 125 and ovarian tumors more than 10 cm in size. They also found that FIGO stage 1a had better survival than those with advanced disease. Complete resection with advanced disease followed by adjuvant chemotherapy was seen to be associated with higher survival.

A case series and review of literature was published in European Oncology and Hematology 2013. [11] In this retrospective review conducted over 24 years, between 1986 and 2010, they found six women at stage III/IV treated for SCC in MCT. In those patients long lasting responses were difficult to achieve but best treatment response was seen in a women who had partial response to chemo-radiotherapy (survival 19 months). According to them, concurrent chemo-radiation could be considered for disease confined to pelvis. However, median survival in their series was only 12.5 months. These reports are similar to our case. While prognosis seems highly dependent on surgical stage, there is a lack of consensus in the literature regarding adjuvant treatment. Platinum based chemotherapy with pelvic radiation may be a reasonable adjuvant therapy for early stage disease.

Conclusion-

Keeping in view the rarity and poor prognosis of SCC of ovary arising in a MCT, it is very essential for a gynaecologic oncologist to be aware of this condition and be equipped to deal with it. The oncopathologists should also always keep it as a differential diagnosis while dealing with dermoid cysts of ovary especially in older females with large sized tumors. Hence, all dermoid cysts should be subjected to histopathological examination and samples must be taken from solid areas to rule out malignant transformation in a mature cystic teratoma.

Legends:

Fig.1a-Gross photograph of right ovarian cystic mass with poltaceous material

Fig.1b-Gross photograph of left ruptured solid and cystic mass with necrotic areas.

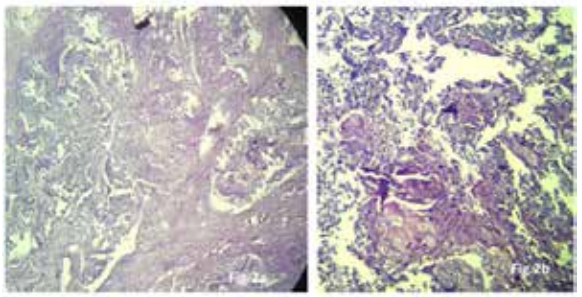

Fig. 2a-Photomicrograph of left ovarian mass showing benign glandular structures with nests of malignant squamous cells.

Fig.2b-Photomicrograph showing malignant squamous cells in nests.

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