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Case Report

Coexistence Of Squamous Cell CarcinomaWith Dermoid Cyst Of Ovary:A Case Report

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Abstract

Ovarian squamous cell carcinoma is a rare malignancy and the occurrence is attributable to malignant transformation of an existing ovarian dermoid cyst. The de novo occurrence of squamous cell carcinoma of the ovary, in the absence of an antecedent ovarian dermoid, is extremely rare. A 40- years- old female presented with pain and enlargement of abdomen since last 4 months. A mass was felt over right iliac fossa. Ultrasonography showed partly cystic and solid area in mass arising from ovary. A tooth- like structure was also seen. Dermoid cyst was the diagnosis offered. Laparotomy was performed. Ovarian mass with hair, a tooth and putty-like material was present. Solid white area with thickened wall was found. Histopathology confirmed the findings of dermoid cyst and the thickened area showed squamous cell carcinoma with areas showing keratinous and giant cell reaction which was also seen at the periphery of tumor areas. Left ovary was normal and no deposits of tumor were seen. Postoperative period was uneventful.

Keywords: Dermoid cyst, Ovary, Squamous cell carcinoma

1. Introduction

Benign cystic teratoma (dermoid cyst) contains tissues that develop from ectoderm, endoderm and mesoderm in an organized manner. 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]. 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. The de novo development of a primary squamous carcinoma, in an otherwise healthy ovary is an extremely rare occurrence[2]. 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.

2. Case Report

A 40- years- old woman, para 3 with 3 living children was admitted in department of Obstetrics and Gynaecology of a rural medical college of central India with complaints of pain and gradual distention of abdomen since 4 months. There was no history of loss of appetite, loss of weight and no bladder or bowel symptoms. On examination, abdomen was soft

non tender with a palpable mass of around 18 weeks pregnant uterus which was hard in consistency. Per vaginal examination revealed uterus as retroverted, retroflexed normal in size with fullness of the right fornix where a nodular mass of size 18 weeks with, variegated consistency was felt. Ultrasonography revealed a large cystic lesion of 12.2x8.1x8.1 cm with solid component of size 5.3x3,9x2.5 cm within right ovary. Preoperative clinical diagnosis of dermoid cyst was rendered. Hysterectomy with bilateral salpingo-oophorectomy was performed. Post operative period was totally uneventful. On gross examination the uterus, cervix and left ovary were normal. Right ovary was enlarged, measuring 13x9x8 cms. External surface was irregular. 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. Microscopic examination (Figure 1, 2) 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. So final diagnosis of dermoid cyst with squamous cell carcinoma was concluded.

Post-operatively, the patient was put on adjuvant chemotherapy with Paclitaxel (175 mg/m^2) and Cisplatin (50 mg/m^2) for 6 cycles. The patient did well after surgery and tolerated 6 cycles of chemotherapy without problems. The patient is under regular follow up and in good condition.

Fig. 1. Microscopic appearance of dermoid cyst with squamous cell carcinoma.

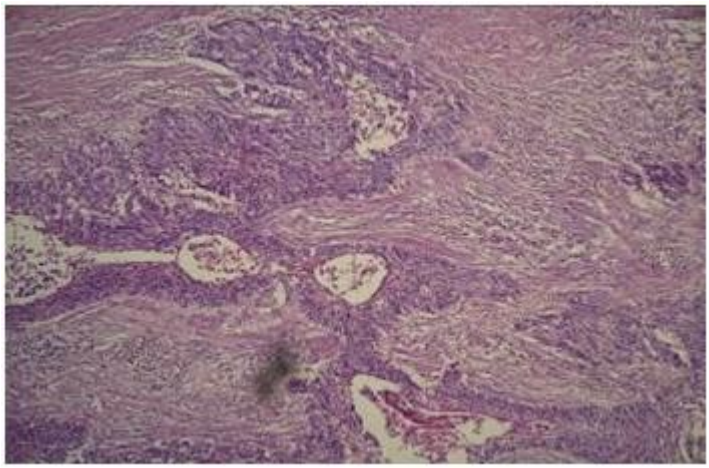
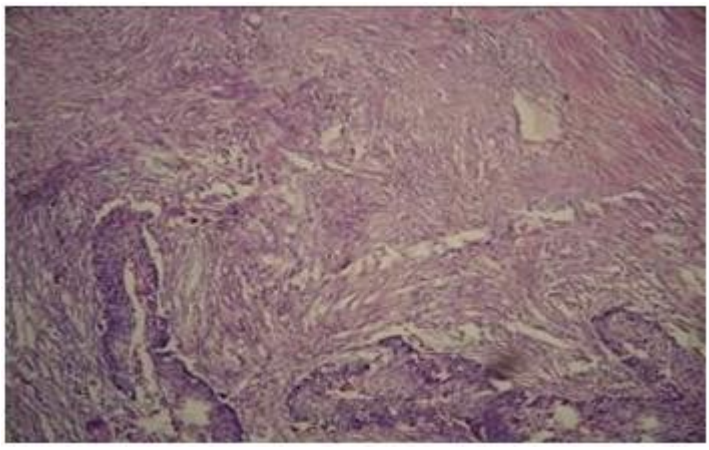


Fig. 2. Microscopic appearance of dermoid cyst with squamous cell carcinoma.



3. Discussion

Ovarian cancer remains the second most common female genital tract malignancy. Mature cystic teratomas, commonly known as dermoids, represent over 10% of the ovarian tumours[1]. On rare occasions (1-2%) the dermoids provide

a background for malignant transformation within components of the teratoma; the majority of such malignancies arising within an ovarian dermoid (80-90%) are squamous cell carcinomas[1]. Malignant transformations have also been reported in endometriotic ovaries. Reports of primary squamous cell carcinomas arising in otherwise healthy ovaries are markedly rare[2], such an occurrence has not previously been described in a patient of Southeast Asian heritage.

Squamous cell carcinoma (SCC) arising in dermoid cysts most probably develops from epidermal elements (80%)[3], although an origin from bronchial epithelium is a possibility. 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]. 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[3]. 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 dermoid cyst is extremely difficult, poses a great challenge and dilemma regarding a need for surgical staging and adjuvant therapy[1]. Risk factors for malignancy in dermoid cyst include patient age, tumor size, imaging characteristics and serum tumor markers. It has been observed that compared to benign dermoid cyst, 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]. 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* [7], 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* [7] 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 dermoid cyst 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 dermoid cyst and SCC in dermoid cyst. In yet, another study Mori *et al* [8] 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 and this includes Computerized tomography with findings of, adnexal mass with fat, calcification, soft tissue component and areas of invasion through the teratoma wall which must raise a suspicion of associated malignancy. Another is Magnetic resonance imaging, which 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.

Due to the rarity of the condition, as well as an incidental nature of the diagnosis, the optimal management approach to primary ovarian squamous cell carcinomas remains unclear. Reported literature suggests that the surgical management for the described cases is akin to that for adenocarcinomas of the ovary, entailing performance of total abdominal hysterectomy, bilateral salpingooophorectomy and omentectomy, with additional steps as needed to ensure surgical debulking of all grossly visible disease. Indeed, optimal cytoreduction and the surgical stage at presentation has been correlated with a significantly improved survival for squamous cell carcinomas arising from dermoid cysts[9]. The role of adjuvant therapy in the management of ovarian squamous cell carcinomas is similarly unclear. Limited experience suggests that surgical management with close follow up alone would suffice for early stage 1A disease[10]. Stages 1C and above have been targeted with a variety of adjuvant regimens with variable outcomes. Relatively poor disease control has been noted with the conventional regimens to treat epithelial ovarian cancers, i.e cisplatin, vincristine, mitomycin C, and bleomycin (POMB)[11]. On the other hand remarkable responses have been achieved with paclitaxel based regimens in some cases[11]. Radiotherapy has also been used with the rationale of squamous cell carcinoma being a radiosensitive tumour; and has brought forth variable results for cancers within antecedent dermoids[1]. In the latest case series and review of literature[1], albeit in cases of squamous cell cancers arising in mature cystic teratoma of the ovary, whole-pelvis radiation and concurrent weekly platinum-based chemotherapy following aggressive cytoreduction has been shown to be of benefit. It however remains unclear whether patients with de novo primary squamous cell cancer of the ovary would benefit as much from similar adjuvant therapy.

4. Conclusion

Primary squamous cell cancers arising in the ovary is an extremely rare entity, and has not previously been described in patients of Southeast Asian heritage. In the absence of well designed trials, and based on the existing albeit sparse data, the primary management approach at present is surgical debulking, akin to the principles underlying the management of ovarian adenocarcinoma. Combination chemotherapy with newer drugs has shown some benefit and pelvic radiotherapy may have a role to play for local control. However, in the absence of quality data, except for the very early stages of presentation, a role for adjuvant therapy is at present unclear.

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