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Pathology

OVARIAN MATURE CYSTIC TERATOMA WITH TRANSFORMATION INTO SQUAMOUS CELL CARCINOMA: REPORT OF TWO CASES

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ABSTRACT Ovarian mature cystic teratomas are the commonest germ cell tumors of the ovary. Malignant transformation in them is a rare event and is difficult to diagnose preoperatively. Clinicians, pathologists and radiologists should always suspect this if the patient is elderly or when the cyst is large or in presence of a solid foci. Here, we discuss two cases of squamous cell carcinoma arising from a mature cystic teratoma.

KEYWORDS : malignant mature cystic teratoma, squamous cell carcinoma, dermoid cyst

INTRODUCTION

Ovarian dermoid cysts also known as Mature cystic teratomas (MCT) are the commonest germ cell tumors of the ovary, characterized pathologically by the presence of elements from at least two of three germ cell layers; ectoderm, endoderm and mesoderm[1,2]. Over 80% of these tumors, which accounts for 20 to 25% of all the ovarian tumors, occur mainly during the reproductive years and may occur in 10 to 20% of women during their lifetime[2,3]. Malignant transformation in these tumors is rare, squamous cell carcinoma (SCC) is the frequent malignancy arising from the ectodermal component of MCT followed by adenocarcinomas [4,5,6].

Here we report two cases of dermoid cyst with malignant transformation because of its rarity and elucidate the importance of these tumors as differentials.

CASE 1

A 60-year-old postmenopausal female presented with complaints of lower abdominal pain and abdominal fullness for 6 months. She had no constitutional symptoms such as loss of appetite, loss of weight with no other significant clinical or family history.

Physical examination revealed a swelling in the left iliac region measuring approximately 12x10 cm. Her blood counts, liver function tests, urea, blood sugar, serum electrolytes, blood cultures and mid-stream urine were normal. Abdominal ultrasound and CT scan revealed a solid cystic mass measuring 12x1x6.5 cm, with areas of calcification in the left pelvic region. Tumors markers such as HCG, CEA, AFP were all within the normal range. The provisional diagnosis thought was a mature cystic teratoma of the left ovary. The patient underwent a total abdominal hysterectomy with bilateral salpingo– oophorectomy. The specimen was sent for histopathologic examination (HPE).

Grossly, an ovarian cyst with smooth, uneven external surface measuring 12 x 11 x 7 cm was identified. Cut section showed a unilocular cyst containing pultaceous material, hair, teeth with a solid grey white papillary structure measuring 2 x 2 cm (Figure 1). Uterus, cervix with right side ovary tube were unremarkable grossly.

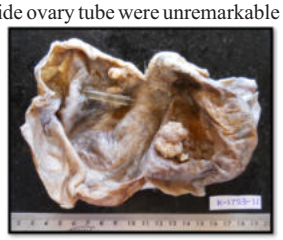

Figure 1: Gross photograph showing cut section of the unilocular cyst with solid grey-white mass measuring 2x2 cm.

Multiple sections from the cystic and solid area revealed a cyst lined by stratified squamous epithelium with nests and sheets of malignant squamous cells in the underlying stroma. These tumour cells are highly pleomorphic having hyperchromatic nuclei with scant amount of cytoplasm, atypical mitotic figures were also noted (Figure 2). The diagnosis of mature cystic teratoma transforming into poorly differentiated squamous cell carcinoma was made. Adjuvant chemotherapy was given to the patient, on follow up after 6 months of treatment her ultrasound and CT scan were normal.

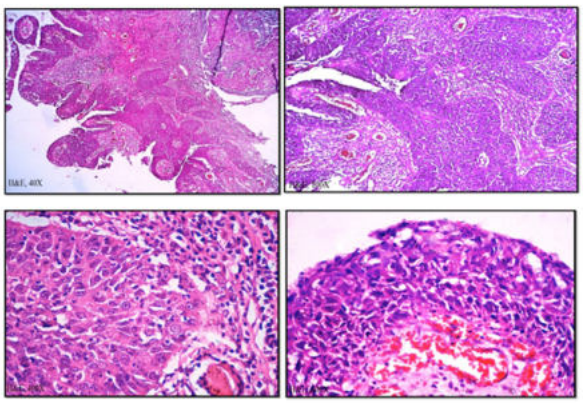

Figure 2: Microscopic photographs showing a cyst lined by stratified squamous epithelium with sheets and nests of malignant squamous cells in the underlying stroma. The tumor cells are highly pleomorphic with hyperchromatic nuclei and scant cytoplasm. Numerous mitotic figures also seen.

CASE 2

A 55-year-old female presented to the emergency department with progressively worsening lower abdominal pain and vomiting. This was severe in nature, predominant in the right pelvic region lasting for a few seconds and resolving spontaneously.

On physical examination she was afebrile, dehydrated with right iliac region tenderness with no rigidity or guarding. Pelvic examination revealed a tender, cystic fixed pelvic mass extending to the right pelvic side wall. Ultrasound scan revealed a right adnexal solid-cystic mass measuring 18 x 11 cm with areas of calcification. Her routine investigations showed microcytic hypochromic anaemia with leucocytosis (13 $X10^9$/L). All the tumour markers (CEA, AFP, HCG) were all within the normal range. The clinical diagnosis thought at this time was mature cystic teratoma of right ovary. Exploratory laparotomy was done and the specimen was sent for histopathological examination.

Grossly an ovarian cyst, weighing 1400gms and measuring 18 x 12 x 8 cm was identified. The external surface was smooth and glistening, cut section showed a unilocular cyst containing pultaceous material and tuft of hair with an 8 x 4 x 2cm grey-white solid area (Figure 3).

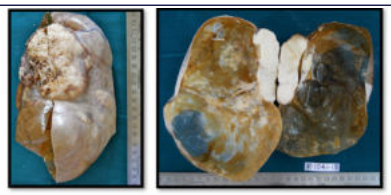

Figure 3: Gross phtograph of an ovarian cyst, cut section showing a unilocular cyst along solid grey-white area measuring 8x4x2 cm.

Multiple sectioning was done and microscopic examination revealed a cyst lined by stratified squamous epithelium showing invasion, with sheets and cords of malignant squamous cells in the underlying stroma. The tumour cells were moderately pleomorphic with vesicular to hyperchromatic nuclei, prominent nucleoli and moderate amount of eosinophilic cytoplasm (Figure 4). Based on these histopathological findings a diagnosis of mature cystic teratoma with malignant transformation into well differentiated squamous cell carcinoma was made. After the first cycle of chemotherapy, she was lost to follow-up.

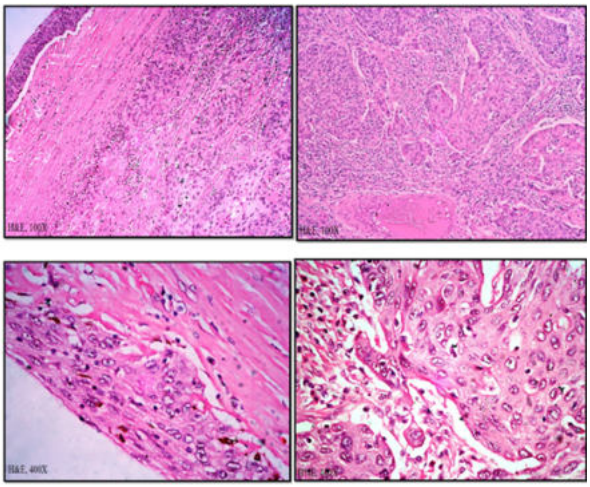

Figure 4: Microphotographs showing a cyst lined by stratified squamous epithelium witl cords and nests of malignant squamous cells in the underlying stroma. The tumor cells a pleomorphic having vesicular nuclei, prominent nucleoli with moderate amount of eosinophilic cytoplasm.

DISCUSSION

Mature cystic teratoma or dermoid cyst is the most common ovarian germ cell neoplasm and the most common childhood ovarian tumor comprising approximately 20 to 25% of all ovarian tumors [5,7].

In different publications the frequency of the malignant change in MCT varies from 1 to 2% irrespective of the age of the patient[1,2,3,4,7,8]. *Kim et al* conducted a study on 560 patients who underwent surgery for a mature cystic teratoma at their hospital and concluded the rate of transformation to be 0.6%[2], similarly *Comerci et al* in 1994 conducted a study on 517 cases and concluded the incidence of malignancy in MCT to as low as 0.17%[6].

Approximately, 75% of malignancies arising in cystic teratomas are invasive or rarely in situ squamous cell carcinomas (SCCs), followed by adenocarcinoma (7%) and sarcoma (7%)[7]. SCC arising in MCTs has been observed in a relatively older patient population although it has been reported in patients as young as 19 years old [8]. The median age at diagnosis of malignant transformation of MCT is 54-61.5 years[9] compared to 37.5 years in MCT [6,7], similar findings were present in our cases.

The patients are usually asymptomatic; although symptoms like abdominal pain and abdomino– pelvic mass may be seen, similar to our cases. It may be discovered accidentally during gynaecologic examination due to mass effect [6,7].Preoperative diagnosis of MCT of ovary is relatively easy due to the radiological detection of bony tissues, cartilage and teeth. However, preoperative detection of malignant transformation of MCTs is challenging due to nonspecific tumor markers and imaging findings. The CT appearance of a MCT is typically fat attenuation within a cyst with, or without, calcification within the wall. In 56 % of the cases, teeth, and other calcified structures have been reported. In malignant transformation there is an irregular border or local tissue invasion[4]

Risk factors for malignancy in a mature cystic teratoma include age over 45 years, tumor diameter greater than 10cm, and rapid growth[5]. *Kikkawa et al*. reported that a tumor diameter of > 9.9 cm was 86% sensitive for malignancy in their series, similar results were documented by *Yamanaka et al*. The utility of tumor markers has also been investigated by multiple studies, *Kikkawa et al* identified that carcinoembryonic antigen (CEA) and squamous cell carcinoma antigen (SCCA) are more sensitive and specific than CA 125. They concluded that a good clinical strategy would be to examine SCCA and CEA levels in patients over 45 years or if the tumour is more than 99 mm in the greatest diameter [11]. According to *Suzuki et al*, SCCA levels were highly dependent on size, that is it is elevated in larger tumors, whether benign or malignant. *Tseng et al*. found that the SCCA level were also useful in detecting relapse [4,10]. However, the optimal level of these tumor markers is also topic of debate[8]

Irrespective of the tumor type, or size of the tumor, prognosis is good in tumors limited to one ovary and with an intact capsule[9]. *Chen at al* in 2008 concluded that the overall 5-year survival rate for all stages is 48.4% and individually in Stage I, II, III and IV it is 75.7%, 33.8%, 20.6% and 0% respectively[12]. The treatment of choice for malignant change in MCT is complete surgical excision, in a nulliparous or young woman it is advisable to perform a unilateral oophorectomy, especially in stage IA disease; however, in the postmenopausal women the surgery of choice would be a total abdominal hysterectomy with bilateral oophorectomy[6]. Postoperative treatment modalities include chemotherapy (single or combined), radiotherapy, or a combination of them. However, due to rarity of this entity patients are unable to participate in large randomized trials, thus the adequate outcome of these treatment modalities is yet to be established [6,9]

CONCLUSION

The pathologist should be always be aware of this possibility in a case of MCT in older women or of large size. Vigilant grossing and sampling need to be done as the malignant component of the tumor might be present in only part of the lesion causing difficulty in suspecting the malignancy.

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