INTRODUCTION: The term teratoma is derived from the Greek word “teratos” which means monster.[1] The Mature Cystic Teratoma (MCT) is the most common ovarian germ cell tumour of the ovary in young women and accounts for 10-20 % of all ovarian tumours.[2] Malignancy in association with benign cystic teratoma of the ovary is rare. Malignancy complicates about 1-2% of dermoid cysts and may occur either by malignant transformation from one of the pre-existing benign elements or a malignant lesion may co-exist with the benign teratoma.[3,4] An incidence of malignancy of 1.8% in 8000 cases of dermoid cysts was found in an extensive study done by Peterson.[5] According to the National Cancer Registry Programme (NCRP) in Manipur, a total of 315 cases of primary malignant ovarian tumour were encountered during the period between January 2005 to December 2012 and only 2 cases (0.63%) were histologically proven to be malignant transformation of mature cystic teratoma. The most common histologic types are squamous cell carcinoma (SCC) followed by adenocarcinoma and melanoma.[4] Malignant transformation occurring in mature cystic teratoma of the ovary is rarely diagnosed pre-operatively due to the rarity of this tumour and its similarity to mature cystic teratoma.[6] Hence, malignant transformation is currently diagnosed by post-operative pathological examination in most cases. In this article, we report two cases of squamous cell carcinoma arising in MCT. The frequency of this type of malignant transformation is age related and is most common in the fifth and sixth decades of life.

CASE REPORTS:

CASE 1: A 71 year old multiparous female, presented with abdominal lump for 2 months and pain abdomen for 5 days. Physical examination revealed a palpable mass in the pelvic region. Routine serological tests and plain abdominal X-ray film revealed no abnormality. Ultrasonography (USG) showed a cystic lesion in the right ovary with a focal small solid area corresponding to an ovarian dermoid cyst. She underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy. Macroscopically, the right ovarian mass measured 18 cm × 18 cm × 10 cm and weighed 1.75kgs. The outer surface was bosselated with many small irregular nodules. On cut section, it was multiloculated and contained pultaceous materials, sebum and hairs and revealed a focal solid area. Uterus, bilateral fallopian tubes and left ovary were grossly unremarkable. On histopathological examination (HPE), sections from the cystic right ovary revealed ectodermal and endodermal derivatives like stratified squamous epithelium, lobules of sebaceous glands, sweat glands, keratin flakes (Figure 1) along with intestinal and respiratory...
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epithelium [Figure 2]. Sections from the solid areas exhibited well differentiated squamous cell carcinoma infiltrating into stroma [Figure 3]. A diagnosis of invasive well differentiated squamous cell carcinoma arising in a mature cystic teratoma was made. Patient was alive and well upto six months. Then she was lost to follow up.

CASE 2: A 55 year-old female presented with abdominal pain and swelling with pre-operative ultrasonography demonstrating features of ovarian dermoid cyst and ascites. Subtotal abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. On gross examination, the uterus, right ovary and bilateral fallopian tubes were unremarkable. The left ovary measured 13 cm ×12 cm ×10 cm and was cystic, biloculated containing pultaceous material and tufts of hair. The cystic wall was greyish-white and thickened focally. On histopathological examinations, sections from the cystic left ovary showed mesodermal and ectodermal derivatives like cartilage, stratified squamous epithelium and sweat glands respectively. The thickened area revealed well differentiated squamous cell carcinoma infiltrating into the stroma. A diagnosis of well differentiated squamous cell carcinoma in a pre-existing mature cystic teratoma was made.

DISCUSSION: Malignant transformation in mature cystic teratoma is a rare pathologic event and occurs in approximately 2% of the cases.[4,5] There is lack of real data regarding the characteristics of these tumours, though there are few articles which gives us some light on these matters. It is of utmost importance to attempt the diagnosis of malignant transformation of MCT pre-operatively as management of ovarian tumours vary depending on the benignity or malignancy. There are no signs or symptoms which are characteristics of malignancy arising in a dermoid cyst. The common symptoms are abdominal pain followed by abdominal mass, but the patient may be asymptomatic or have symptoms of abdominal distension or bloated abdomen as caused by benign cysts.[6]

The diagnosis of MCT by radiologic means is possible due the presence of cysts, bones, teeth and cartilages. However, pre-operative diagnosis of malignant transformation can be very challenging as the features can be similar to uncomplicated MCT. Hence, malignant transformation should be in our mind while considering MCT.

A suitable marker for malignant transformation in MCT is considered by the presence of both the following parameters viz. patients age above 40 years and high serum squamous cell carcinoma (SCC) antigen level above 2.5ng/ml.[6,7] Accordingly, urgent surgical treatment may be considered when there is high pre-operative serum level of SCC antigen. SCC was commonly observed in relatively older patient and the cut off value was 45 years.[2] The age of the patients in our study is 71 and 55 years and this is consistent with other studies done earlier.[1,3,4,5,6] A regular USG of the pelvis in elderly patient may help in early diagnosis of this type of tumour.

The risk of tumour is increased with increasing tumour size. It was reported that a tumour diameter of larger than 9.9 cm was 86% sensitive for malignancy in a series.[2] The mean size of the tumour in our study was 15.2 cms which correlate with other studies.[1-9] Hence, an ovarian tumour with size > 10 cms should arouse our mind for the possibility of malignant transformation of MCT.
Histologically, a poor prognosis is associated with SCC arising in MCT. Kikkawa et al were the first to correlate prognosis of the patient with the type of stromal invasion. Mode of stromal invasion is classified into 3 types – alpha, beta and gamma modes. In alpha mode, the tumour cells invades the stroma with a well-defined border between tumour and stroma, while in gamma mode there is no clear demarcation between tumour and stroma. Beta mode shows intermediate between alpha and gamma. Our patients were two postmenopausal females having large ovarian mass with solid areas which on histopathological section showed features of malignant squamous cells with alpha mode of invasion.

In conclusion, this uncommon tumour should always be on the back of our mind while encountering a large ovarian tumour in an elderly patient. Different treatment options should be carefully considered, should the tumour turn out to be malignant.

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Fig. 1: HPE of cystic ovary showing ectodermal derivatives like stratified squamous epithelium, lobules of sebaceous glands and keratin flakes. H & E; ×400

Fig. 2: HPE of cystic wall showing respiratory lining epithelium. H&E; ×400

Fig. 3: HPE from the solid area of the ovarian mass showing well differentiated squamous cell carcinoma. H&E; ×400
**AUTHORS:**
1. Babina Sorokhaibam
2. Khurajam Sucheta Devi
3. Sushma Khurajam
4. Deepak Laishram
5. Seter Potom

**PARTICULARS OF CONTRIBUTORS:**
1. Associate Professor, Department of Pathology, Regional Institute of Medical Sciences.
2. Assistant Professor, Department of Pathology, Regional Institute of Medical Sciences.
3. Associate Professor, Department of Pathology, Regional Institute of Medical Sciences.
4. Trainee, Department of Pathology, Regional Institute of Medical Sciences.
5. Trainee, Department of Pathology, Regional Institute of Medical Sciences.

**NAME ADDRESS EMAIL ID OF THE CORRESPONDING AUTHOR:**
Dr. Khurajam Sucheta Devi,
Department of Pathology,
Regional Institute of Medical Sciences,
Lamphel, Imphal-795004,
Manipur, India.
E-mail: sucheta.kh73@gmail.com

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