Case Report

Mature cystic teratoma with malignant transformation of teratomatous urothelial cells: Rare case presentation

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ABSTRACT

The occurrence of malignancies in somatic elements of mature cystic teratoma of ovary is rare. The malignancies that may be encountered in dermoid cyst include squamous cell carcinoma, adenocarcinoma, adenosquamous carcinoma, melanoma, sarcoma, carcinoid, and germ cell neoplasms. The development of transitional cell carcinoma (TCC) in dermoid cyst is extremely rare with only four such cases having been reported in literature so far. Here we report the fifth case of such an occurrence in a 50-year-old postmenopausal multiparous female patient. She presented with pain and gradual swelling of abdomen for 1 month. Abdominal computed tomography revealed a solid space occupying lesion with few cystic components at right pelvis, raising the possibility of an ovarian neoplasm. The level of CA-125 was slightly raised (56·45 U/ml). Total abdominal hysterectomy and bilateral salpingo-oopherectomy was performed. Microscopic examination showed cyst wall lined by stratified squamous epithelium. Beneath the cyst wall, a tumor mass was present, histological features of which resembled that of high-grade TCC (stage pT1aNXMX). On immunohistochemical analysis, the tumor was found to be positive for CK7 and CK20 and negative for WT-1. These results were consistent with a diagnosis of TCC arising in urothelium of mature cystic teratoma. Reporting of such extremely rare cases is important for the assessment of prognostic factors and treatment protocols.

Key words: Benign cystic teratoma, immunohistochemistry, transitional cell carcinoma

INTRODUCTION

Mature cystic teratoma is one of the most common ovarian neoplasms that occurs in women of reproductive age group. It constitutes nearly 95% of all ovarian tumors of germ cell origin.[1] However, the presence of malignant somatic elements within benign cystic teratoma is a rare event, accounting for approximately 1% of cases.[2] Squamous cell carcinoma is the most commonly found malignancy in mature cystic teratoma, followed by adenocarcinoma and carcinoid tumor.[3] Here, we report a case of transitional cell carcinoma (TCC) arising in benign cystic teratoma, which is an extremely rare occurrence.

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CASE REPORT

A postmenopausal nondiabetic 50-year-old female patient presented with pain and gradual swelling of abdomen for the last 1 month. She was multiparous (P_{5+0}).

General examination revealed that the patient had pallor and stable vitals. Abdominal examination revealed the presence of a large nontender swelling, approximately 12 cm × 9 cm in dimensions, involving the right iliac and hypogastric regions. There was no ascites or hepatosplenomegaly. Per vaginal examination revealed that the uterus was normal in size, mobile, with clear fornices.

Routine hematological tests showed that the hemoglobin level of the patient was 10.5 g/dl and her total and differential counts were within normal limits.

Abdominal computed tomography (CT) was done which revealed a solid space occupying lesion with few cystic components at right pelvis. The lesion was approximately

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11 cm × 8 cm in dimensions and had irregular margins. On the basis of the radiologic findings, the lesion was suspected to be a right ovarian neoplasm [Figure 1a].

An assay for CA-125 was advised. The level of CA-125 was slightly raised (56·45 U/ml).

Laparotomy was undertaken for removal of the mass. When the abdomen was opened, no peritoneal fluid was seen. Peritoneal wash was given with normal saline, and the washing was sent for cytologic examination. A large mass, 11 cm × 10 cm in size, with bosselated surface, was found in relation to the right ovary. Total abdominal hysterectomy and bilateral salpingo-oophorectomy was performed.

Cytologic examination of the fluid did not reveal any malignant cells. Gross examination of the resected specimen revealed a large tumor mass arising from the right ovary, measuring approximately 11 cm × 9 cm × 6 cm. The outer surface of the mass was smooth, without any capsular breach. Cut surface showed both solid and cystic areas. Pultaceous material was found within the mass [Figure 1b].

Microscopic examination showed cyst wall lined by stratified squamous epithelium. The wall contained benign teratomatous structures such as skin adnexae, thyroid follicles, and mature adipose tissue. Deep to the cyst wall, the presence of tumor mass was noted, composed of pleomorphic urothelial cells arranged in papillae and cell nests, resembling that of high-grade TCC. The tumor was diagnosed as mature cystic teratoma of ovary with TCC (stage pT1aNXMX) [Figure 2a and b].

Immunohistochemical analysis was performed, and the transitional cell carcinomatous component was found to be positive for CK7 and CK20. This component, however, gave negative results with WT-1. These results were consistent with a diagnosis of TCC arising in urothelium of mature cystic teratoma [Figure 2c and d].

The patient has been on follow-up for the last 1 year which was uneventful.

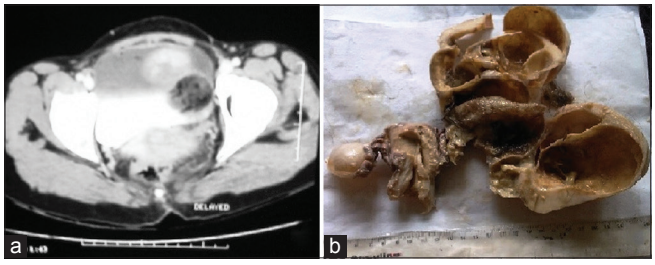

Figure 1: (a) Computed tomography scan picture showing a solid space occupying lesion with few cystic components at right pelvis, (b) gross picture of resected specimen – cut surface of right adnexal mass revealed both cystic and solid areas

DISCUSSION

The commonly encountered malignancies in mature cystic teratoma of ovary include squamous cell carcinoma, adenocarcinoma, adenosquamous carcinoma, melanoma, sarcoma, carcinoid, and germ cell neoplasms.[2] The occurrence of TCC in benign cystic teratoma of ovary is an extremely rare event with only four such cases having been reported in literature so far.[2,4-6]

The pathogenesis of malignant transformation in dermoid cyst is unclear. It has been postulated that prolonged irritation of teratomatous urothelium within dermoid cyst may lead to the development of urothelial carcinoma.[6]

Malignant transformation in benign cystic teratoma is suspected when the following factors are present: Age above 45 years, tumor diameter exceeding 10 cm, sudden increase in size and characteristic imaging findings.[2] All these factors were present in this case. However, a high index of suspicion is always recommended because malignancy in dermoid cyst has been reported in patients as young as 19 years.[7]

The common diagnostic modalities include ultrasound, CT scan, and serologic markers.[8] The level of CA-125 has been found to be of use in differentiating benign cystic teratoma from malignancy in dermoid cyst (17 U/ml vs. 33 U/ml).[6] In this case, the CA-125 level was 56 · 45 U/ml.

Lee and Lee stated that TCC arising in mature cystic teratoma must be distinguished from primary ovarian TCC, which is a surface epithelial tumor.[6] This may be done on the basis of immunohistochemical analysis. TCC arising

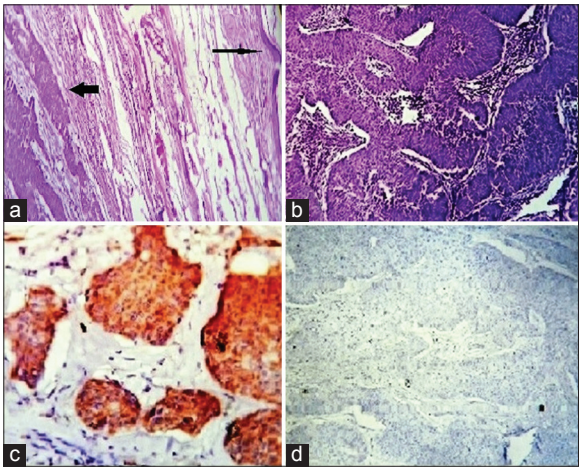

Figure 2: (a) Photomicrograph showing cyst wall lined by stratified squamous epithelium (thin arrow), with areas of transitional cell carcinoma (TCC) (thick arrow) beneath (H and E, ×40), (b) TCC-pleomorphic urothelial cells arranged in papillae and cell nests (H and E, ×100), (c) CK20 positive TCC cells (×400), (d) WT-1 negative TCC cells (×100)

from urothelium is positive for CK7, CK20, uroplakin III, thrombomodulin, and negative for WT-1. On the other hand, primary ovarian TCC yields positive results with WT-1 and CK7. In nearly 80% cases, primary TCC of ovary is negative for CK20, uroplakin III, and thrombomodulin.[1]

In the present case, TCC detected within mature cystic teratoma was positive for both CK7 and CK20. The tumor was negative for WT-1. These results showed that the TCC originated from urothelium of mature cytic teratoma and that it was not a surface epithelial tumor of ovarian origin. The possibility of ovarian metastasis from carcinoma of urinary tract was ruled out when a thorough clinical check-up and radiological investigations did not reveal any abnormality of the urinary system. Hence, a diagnosis of TCC arising in mature cystic teratoma was rendered.

Due to the rarity of malignancies in dermoid cysts, prognostic parameters and treatment modalities of such cases remain controversial. Most authors agree that age of patient, tumor size, and stage of the disease are the important factors that influence survival.[2]

Management usually consists of total abdominal hysterectomy and bilateral salpingo-oophorectomy, with or without adjuvant chemotherapy.[5,6] In advanced stage disease, combination chemotherapy with platinum/taxane has been found to be effective.[9]

CONCLUSION

The occurrence of TCC in dermoid cyst is rare and to the best of our knowledge, this is the fifth such case being reported in the literature. Determination of prognostic factors and standardization of treatment protocols require immediate reporting of secondary malignancies in mature cystic teratomas. Retrospective studies from institutes with large databases are necessary for better understanding of such rare malignancies.

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