

Case Report

Ovarian squamous cell carcinoma arising from mature cystic teratoma in a young patient

Belson Rugwizangoga^{1,2*}, Marie-Claire Ndayisaba², Narcisse Niyikora², Vénant Niyikiza³, Louise Kalisa^{1,4}, Vénérand Bigirimana²

¹School of Medicine and Pharmacy, University of Rwanda, P.O.Box 3286 Kigali, Rwanda.

²Department of Laboratory, University Teaching Hospital of Kigali, P.O.Box 655 Kigali, Rwanda.

³Department of Gynaecology and Obstetrics, University Teaching Hospital of Kigali, P.O.Box 655 Kigali, Rwanda.

⁴Department of Radiology, University Teaching Hospital of Kigali, P.O.Box 655 Kigali, Rwanda.

Abstract

Squamous cell carcinoma arising from ovarian mature cystic teratoma (MCT) is very rare. We report a case of a 21-year-old female who presented with a 7-month history of abdominal mass increasing in size with time. Imaging findings were suggestive of bilateral ovarian teratoma. Intraoperative findings were cystic masses in both ovaries and a solid multinodular left mesenteric mass. On gross examination, there were cysts filled with gelatinous material and hairs, while the mesenteric mass consisted of matted lymph nodes. Histology revealed a MCT with multifocal areas of invasive squamous cell carcinoma, in all three masses. While squamous cell carcinoma in MCT is very rare, this is a unique case reported in a so young patient. The younger age could not be a factor precluding malignancy in MCT.

Key words:

Ovary; mature cystic teratoma; squamous cell carcinoma; young patient

Introduction

Squamous cell carcinoma arising from ovarian mature cystic teratoma (MCT) is very rare, and represents only 0.006% of all MCT in large series of study [1,2]. In previous reports, the age at diagnosis ranges from 33years upwards, while the tumour size ranges 9 to16 cm [1,3]. Biochemical tests of squamous cell carcinoma antigen levels have diagnostic and staging value, while clinical and imaging findings usually suggest a benign cyst [1]. This is a case of an extremely rare primary ovarian tumour, and furthermore in an unusual age group; its diagnosis could be done only by histology, while other diagnostic procedures suggested a benign cyst.

Article history:

Received: 26/03/2015

Accepted: 18/06/2015

*Correspondence: Dr. Belson Rugwizangoga, MD, MMed (Pathol)

Address: Department of Clinical Biology

University of Rwanda School of Medicine and Pharmacy/

University Teaching Hospital of Kigali (CHUK) P.O.Box 655 Kigali, Rwanda

Phone: +250788546597

E-mail: belson777@gmail.com

shunt [7]. Risks are particularly high when the average systemic oxygen saturation falls below 80-85% [2].

Case presentation

We report a case of a 21-year-old female who presented with a 7-month history of intermittent abdominal non-irradiating pain, worsening when lying down, and alleviated by painkillers. Two months later, she started filling a mass in the abdomen. She consulted a district hospital where physical examination suspected a tumour of ovary or caecum. The patient was referred to the university teaching hospital 6 months after the onset of the symptoms. Medical and familial history was unremarkable; she had regular menses without amenorrhea. On physical examination, the abdomen was soft, not distended and not tender. There was a hard irregular mass in the hypogastrium extending to the right flank measuring 12x8cm. Haematological tests were normal; blood urea and creatinine were in normal ranges. Chest X-ray film was normal. Computerized tomography (CT) scans showed

right ovarian complex mass with calcifications and cystic component, suggestive of right ovarian dermoid cyst with suspicion of malignant transformation, the differential diagnosis being sarcoma with peritoneal metastasis. The setting has neither interventional radiology experts, nor other advanced imaging techniques other than CT scan.

Figure 1.

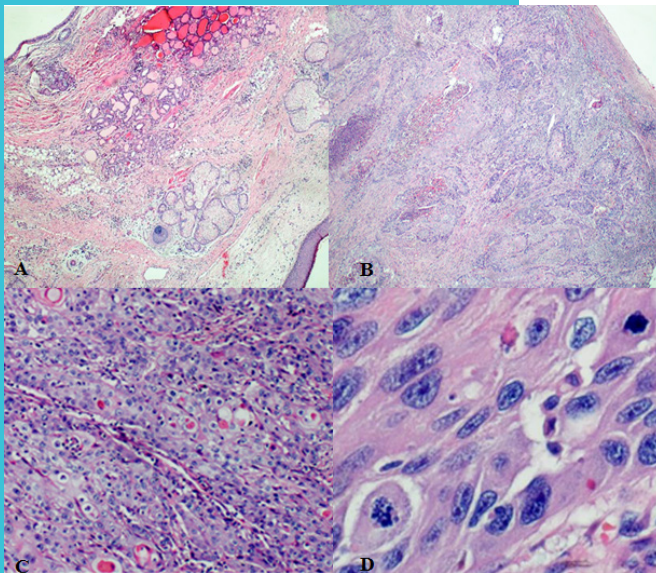


Figure 1. Photomicrographs. *A*, low power photomicrograph showing a mature cystic teratoma component, composed of skin and its appendages (right lower corner to central part), thyroid tissue (upper part) and a respiratory epithelium (left upper corner) (H&E x10). *B*, low power photomicrograph shows a solid trabecular tumour with areas of comedo necrosis; the capsule of the cyst is at right upper corner (H&E x10). *C*, intermediate power photomicrograph exhibits solid nests of large polygonal cells with large cytoplasm, keratinization; large nuclei and a reactive inflammatory infiltrate in the stroma. *D*, high power view of the tumour shows nuclear pleomorphism and hyperchromatism, open chromatin, multiple prominent nucleoli and atypical mitoses.

Tumour markers were not analysed because the hospital had not the capacity of analysing tumour markers other than total prostate-specific antigen (PSA). Vertical laparotomy was performed one day following admission. Intraoperative findings were left ovarian cyst adherent to bowel and omentum, right ovarian mass adherent to the bowel and omentum, and left mesenteric lymphadenopathies. Frozen sections were not performed because the hospital had no cryostat. Excision of both masses and the left mesenteric nodes was performed, the specimens sent for histopathology. The left ovarian cyst wall measured 9x7x3cm; its inner surface was hairy and showed a firm nodule, cartilaginous and cystic on cut sections. The right ovarian mass measured 9x7x4cm and weighed 158 g; it had an irregular surface with a necrotic area; the cut sections were yellowish with areas of haemorrhage and cystic loculations. The third piece weighed 75 g and measured 8x6x2.5cm, consisting of a mesenteric tissue attached to an irregular firm tissue of matted lymph nodes 5x2x1 cm in size; it was solid whitish on cut sections, with cystic formations. Two lymph nodes were isolated in the attached mesentery. Histologically, the left ovarian tissue sections showed a cystic lesion exhibiting skin and its appendages, neural tissue, thyroid, cartilage and salivary glands. The sections of the left ovarian mass, the right ovarian mass, the mesenteric matted nodes and one of the isolate lymph nodes showed a trabecular tumour made up of cohesive large polygonal cells exhibiting enlarged atypical nuclei with frequent multipolar mitoses, large eosinophilic cytoplasm with intercellular bridges and keratin pearls (Figure 1 A-D). The intervening stroma was inflamed. One of the isolate nodes showed reactive follicular hyperplasia. The morphological features were of left ovarian teratoma with Struma ovarii, and an infiltrating squamous cell carcinoma involving left and right ovaries and left mesenteric lymph nodes. The patient was discharged from hospital three days after surgery waiting for the histology results, and was lost-to-follow-up for 9 months because of changing her residence district and phone contacts. This compromised the further treatment options including cytoreductive surgery and chemotherapy. She was retrieved through administrative authorities but died of this disease five days later.

Discussion

The primary ovarian squamous cell carcinoma is extremely rare [1]. In some of these cases, the squamous cell carcinoma arises from a mature cystic teratoma; it may arise from an ovarian endometriosis on the other hand [1,2]. When an ovarian squamous cell carcinoma arises from a mature cystic teratoma, two origins are possible, namely either epidermal or respiratory epithelium [4]. Previous data also exemplified a squamous cell carcinoma in situ arising from a MCT [5]. Squamous cell carcinoma arising from ovarian mature cystic teratoma is very rare, and represents only 0.006% of all MCT in large series of study [1]. The reported age at diagnosis is 33 years and above [1,3]. To the best of our knowledge, this case concerns a patient younger than any other reported cases elsewhere, with a primary ovarian squamous cell carcinoma. This should prompt the revision of the assumption that a clinical diagnosis of ovarian squamous cell carcinoma must be considered when the patient is 45 years of age and above [6]. Different subtypes of squamous cell carcinoma may be encountered in MCT [7]. In the literature, the size of a mass of primary ovarian squamous cell carcinoma varies from 9 to 16 cm, and a MCT with a size of more than 10 cm in greatest dimension has been associated with increased risk of malignancy [1,2,4]. This case had three fragments with each one having about 9 cm in greatest dimension. The fragmentation makes it difficult for comparison to other cases, but this seems to be larger than 16 cm in aggregate. The gross examination on cut sections shows a variety of features, but among them necrosis is an important clue to a malignancy [2]. The same features were present in this case.

Squamous cell carcinoma antigen level has a diagnostic and staging value [1]. In our setting, this biochemical test is not available, and therefore only histology could give the diagnosis and orient further management. As for other tumour markers, the availability of this test and its accessibility to the population should be hastened. The therapeutic option for this patient was tumour excision with left mesenteric lymphadenectomy. The cases of similar advanced stage of disease are treated with surgery and combination chemotherapy [4]. The loss-to-follow-up of this patient has resulted in failure to add combination therapy to the surgery. Squamous cell carcinoma arising in a cystic teratoma is very rare. This is a unique case proving that squamous cell carcinoma arising in a cystic teratoma of the ovary may occur at younger age than was previously documented. The tumour size and imaging findings are important factors in making a differential diagnosis of squamous cell carcinoma in a cystic teratoma of the ovary. Therefore, SCC marker and CEA levels should be measured in patients in whom clinical and/or imaging findings are suspicious of malignancy, independently of the age.

Acknowledgement

None

Conflict of Interest Statement

The authors declare no conflict of interest

Authors' contributions: BR conceived the idea, VN and M-CN analysed the patient's data regarding the disease; BR, M-CN, NN and VB performed the histological examination of the tissue; BR, NN and M-CN did literature search, wrote the article and edited it, VB critically reviewed the article for its final content.

References

1. Choi E-J, Koo Y-J, Jeon J-H, Kim T-J, Lee K-H, Lim K-T. Clinical experience in ovarian squamous cell carcinoma arising from mature cystic teratoma: A rare entity. *Obs Gynecol Sci* 2014;57(4):274–80.
2. Mahe E, Sur M. Primary Squamous Cell Carcinoma of the Ovary. *MUMJ* 2011;8(1):80–3.
3. Bashyal R, Lee M. Squamous cell carcinoma arising in ovarian mature cystic teratoma: report of three cases. *J Pathol Nepal* 2012;2:248–50.
4. Mardi K, Sharma S. Squamous cell carcinoma arising in an ovarian mature cystic teratoma. *Clin Cancer Investig J* 2014;3(1):2014–6.
5. Zakkouri FA, Ouaouch S, Boutayeb S, Rimani M, Gamra L, Mrabti H. Squamous cell carcinoma in situ arising in mature cystic teratoma of the ovary: a case report. *J Ovarian Res* 2011;4(1):5.
6. Kikkawa F, Nawa A, Tamakoshi K, Ishikawa H, Kuzuya K, Suganuma N, et al. Diagnosis of Squamous Cell Carcinoma Arising from Mature Cystic Teratoma of the Ovary. *Cancer* 1998;82:2249–55.
7. Prameela RC, Satyanarayana C. Pseudoglandular (Adenoid, Acantholytic) Squamous Cell Carcinoma in Mature Cystic Teratoma of the Ovary: A Case Report. *Int J Sci Study* 2014;2(8):204–6.