

Bilateral ovarian mature cystic teratoma co-existing with granulosa cell tumour: a case report

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Abstract

Background: Mature granulosa cell tumour (GCT) is a rare neoplasm. it accounts for about 1-2% of all ovarian tumours. Even more infrequent is the coexistence of GCT with mature cystic teratoma in the same ovary.

Case presentation: A 53-year-old woman with constipation, abdominal swelling, and pain presented to our gynaecology clinic. Physical examination revealed a distended abdomen, ascites, cystocele, stress incontinence, and external haemorrhoids. A cystic teratoma co-existing with a granulosa cell tumour was diagnosed histologically. Adjuvant chemotherapy containing Cisplatin [75mg/m²] and Docetaxel [75mg/m²] was completed, followed by radiotherapy. She has been clinically stable for two years and is currently on long-term follow-up to forestall recurrence.

Conclusion: The coexistence of mature granulosa cell tumour (GCT) with mature cystic teratoma especially in the same ovary is an extremely rare pathology. Although total abdominal hysterectomy and bilateral salpingo-oophorectomy are the treatments of choice in postmenopausal and perimenopausal women, these may not always be feasible owing to the nature of the presentation. Adjuvant chemotherapy and radiotherapy may be beneficial as high cases of recurrence have been reported. Follow-ups are strongly advocated due to the recurrence that can occur even as late as 20 years following the removal of the primary neoplasm.

Keywords: Teratoma, Granulosa cell tumour, Ovarian neoplasm, Histology

Introduction

Teratoma remains the most common germ cell tumour of the ovary, accounting for over 20% of all ovarian neoplasms (1). They are broadly classified as follows: mature cystic teratoma, immature teratomas, monodermal teratomas (including struma ovarii, carcinoid tumours, neuroectodermal tumours, and sebaceous tumours), and fetiform teratomas (2). Mature cystic teratoma also referred to as dermoid cysts, is the most common of these, accounting for over

70% of benign tumours in the reproductive years and 20% in postmenopausal women (3). On the other hand, non-germ elements of the ovarian sex-cord stromal cells give rise to ovarian granulosa cell tumours. These represent a rare group of tumours, constituting only about 2-5% of all ovarian cancers (4, 5). They are further classified as adult and juvenile, with the former being more common and predominantly affecting peri-menopausal and post-menopausal women (5). Dermoid cysts coexisting with granulosa cell

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tumours in the same ovary are not common and are considered to represent two synchronously occurring independent tumours (6). We herein discuss a case of mature cystic teratoma co-existing with a granulosa cell tumour in a post-menopausal Nigerian woman.

Case presentation

A 53-year-old para 3⁺⁵ (3 Alive) woman presented to our gynaecology clinic on account of constipation of 3 months duration, progressive abdominal swelling, and pain of one-month duration. She had associated altered bowel habits, the passage of pellet-like stools, significant weight loss, anorexia, nausea, and early satiety. No tenesmus or bleeding per rectum. No family history of breast, colon, or ovarian cancer. She did not smoke cigarettes or drink alcohol. She was 18 months post-menopausal. Her obstetric history included a voluntary termination of pregnancy with two spontaneous terminations of pregnancy.

Physical examination revealed a middle-aged woman, chronically ill-looking, mildly pale, and anicteric with no peripheral lymphadenopathy. She was tachycardic. Her abdomen was grossly distended with difficult palpation of the viscera. Ascites were demonstrable by shifting dullness. A vaginal examination revealed a grade II cystocele with stress incontinence. The cervix was firm, with the os closed. Cervical excitation tenderness was elicited, with a full and tender Pouch of Douglas. Rectal examination revealed external haemorrhoids. There was an extrinsic mass impinging on the anterior rectal wall with a mobile rectal wall.

Abdomino-pelvic ultrasound scan showed a complex right adnexal heterogeneous multiseptated thick-walled mass measuring 12.2x8.0x12.5cm with gross ascites. Contrast-enhanced abdominopelvic CT scan suggested bilateral teratoma, with the right larger than the

left, with peritoneal deposits and moderate ascites. A simple hepatic cyst was also seen. She was anaemic with a PCV of 27.2%. Her alkaline phosphatase (ALP) and gamma-glutamyl phosphatase (GGT) were slightly elevated at 141 IU/L and 95 IU/L respectively.

She was stabilized and transfused while working up for an exploratory laparotomy with bilateral ovariectomy. Intra-op findings included 7 litres of ascitic fluid, a huge right ovarian mass measuring 12x12cm, left ovarian mass measuring 5x4 cm compressing the sigmoid colon. The uterus was 10 weeks in size with multiple tumor nodules in the anterior and posterior walls. The uterus was bound down in the Pouch of Douglas and uterovesical peritoneum. Multiple tumor nodules, >2cm were found on the parenchymal loops of the caecum, ascending colon, and sigmoid colon. Tiny nodules, <1cm (2 sites) in the parietal surface of the liver and falciform ligament were also seen. Hysterectomy was deemed impossible due to extensive tumour nodules binding down the uterus in the Pouch of Douglas and uterovesical region.

In histopathological evaluation, the macroscopy showed right and left nodular and cystic greyish-white ovarian masses measuring 12.0x8.0x6.0cm and 4.0x3.0x2.0cm respectively. Cut surfaces of the cystic masses were similar and showed nodular greyish-white tissue with cysts filled with tufts of hair and areas of bony tissue. Microscopically, sections of the right ovary revealed clusters of granulosa cells forming macrofollicular patterns in a fibroblastic stroma. Most of the follicles appeared empty, and some contained loose fibrous eosinophilic material (Figure 1). Also seen in sections of the right and left ovaries were strands of epithelial cells forming part of cyst-like structures (Figure 2), and clusters of matted adipocytes and areas of calcifications (Figure 3).

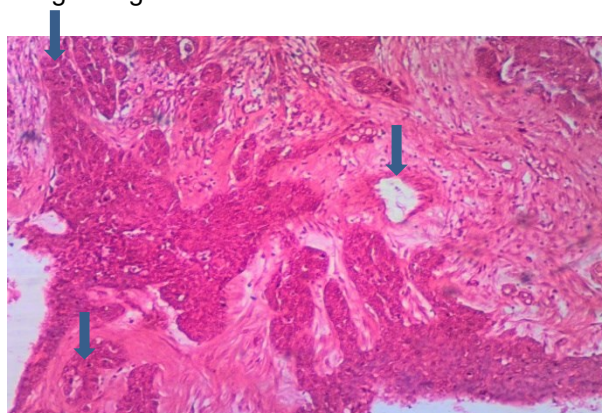


FIGURE 1: (H & E X 40) shows right ovarian tissue with clusters of granulosa cells forming macro-follicles in areas some of which are empty-appearing (arrows) in keeping with a granulosa cell tumour.

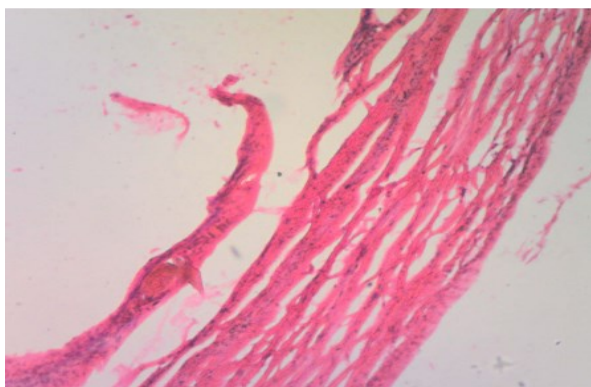


FIGURE 2: (H &E X 40) shows right ovarian tissue with focal areas of strands of flattened epithelial cells forming part of cyst-like structures suggestive of a mature cystic teratoma

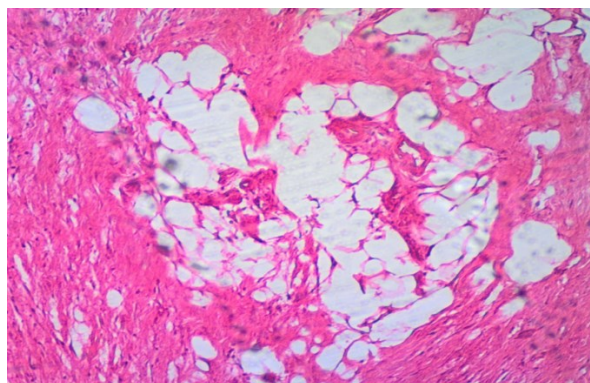


FIGURE 3: (H &E X 100) shows left ovarian tissue with focal clusters of mature adipose tissue in keeping with a mature cystic teratoma

An impression of bilateral mature cystic teratoma with a right ovarian granulosa cell tumour was made following histology. She had 3 units of whole blood intra-op and 2 units post-op. The post-operative period was complicated by paralytic ileus secondary to hypokalemia and was treated as such. She was discharged home 9 days post-op. She was commenced on a Cisplatin (75 mg/m²)/Docetaxel (75 mg/m²) regimen following recuperation due to the unavailability of the BEP (Bleomycin/Etoposide/Cisplatin) regimen. Six cycles of three weeks each of the chemotherapies were given, along with a five-day preventive course of dexamethasone (8 mg, twice daily). She subsequently had radiotherapy in another centre and has been clinically stable 2 years afterwards with no evidence of disease recurrence. We intend to follow up with our patient for at least 20 years post-intervention considering the high recurrence rate of the pathology.

Discussion

Mature cystic teratomas comprise about 20% of all ovarian neoplasms, occur mostly in young individuals (less than 45 years) (1), and are bilateral in about 9% of cases as in our patient (7). The majority of cases of mature cystic teratomas are asymptomatic unless associated

with complications or paraneoplastic syndromes. Complications include torsion (16%), rupture (1-4%), malignant transformation (1-2%), infection (1%), and autoimmune hemolytic anemia (<1%) (8). Ovarian granulosa cell tumours are particularly characterized by their ability to elaborate feminizing hormones and are thus classified as ovarian feminizing mesenchymomas (9).

Unusual clinical presentations of mature cystic teratoma have been reported in the literature, with colorectal involvement being very rare. Rectal wall invasion may present with altered bowel habits due to faeces filling the cyst, hence causing compression of the rectum (10). Our patient presented with local and distant spread/seeding of granulosa cell tumours into the peritoneum and distant sites.

Transvaginal ultrasound scan (TVS) remains the imaging modality of choice in evaluating asymptomatic women presenting with pelvic masses including ovarian mature cystic teratoma (3). Ultrasound features of ovarian mature cystic teratomas include echogenic sebaceous material and calcification, typically containing a hypoechoic attenuating component with multiple small homogeneous interfaces (3). Furthermore, radiological features suggestive of malignant ovarian masses include irregular, thick walls and septations, and papillary projections, with solid,

echogenic foci (11, 12). In our patient, USS revealed a complex right adnexal heterogeneous multiseptated thick-walled mass.

CT scan has a high sensitivity (93-98%) in the evaluation of adnexal masses and is pivotal when ultrasound is non-diagnostic. Additionally, it has a role in demonstrating fat, calcifications, surrounding spread, and mass effect on adjacent structures (13).

Histologically, mature cystic teratomas are usually thick-walled and contain septa dividing them into several compartments, with Rokitansky protuberances (raised protuberance projecting in the cavity). The cysts may contain bone, teeth, and hair as seen in our patient. The cyst wall is usually lined by a hyalinized squamous epithelium. The diagnosis of immature teratoma is made if any immature tissue is seen histologically (7, 13).

The adult subtype of granulosa cell tumours has five histologic patterns including micro follicle, macro follicle, insular, trabecular, and spindle/sarcomatoid patterns. Specifically, microfollicular patterns with Call-Exner bodies and coffee bean nuclei are pathognomonic (14). Controversies exist concerning the best surgical management of ovarian mature cystic teratoma, with a paucity of clinical trials to evaluate the various options (3). Oophorectomy is advocated in postmenopausal and perimenopausal women, particularly with coexisting tumours, multiple cysts, or large teratomas where there is little conservable ovarian tissue left (3). Furthermore, in the case of granulosa cell tumours, total abdominal hysterectomy with bilateral salpingo-oophorectomy is advocated (15, 16). This may be followed up with adjuvant chemotherapy and radiotherapy in extraovarian cases although there is no consensus on this (17). The chemotherapy of choice is bleomycin, etoposide, and cisplatin regimen (18) or carboplatin, paclitaxel regimen (19). These tumours, however, occasionally show poor responses to chemotherapy and are aggressive. In our case, intraoperatively, our patient had bilateral oophorectomy only as a hysterectomy was deemed impossible due to extensive intraperitoneal tumour seedling; this was followed up with adjuvant chemotherapy with cisplatin/docetaxel regimen and radiotherapy.

Conclusion

In conclusion, we report a case of bilateral ovarian mature cystic teratoma coexisting with a granulosa cell tumour. Histological identification is key in its management. Although total abdominal hysterectomy and bilateral salpingo-oophorectomy are the treatments of choice in postmenopausal and perimenopausal women, especially in certain conditions, these may not

always be feasible owing to the nature of the presentation. Long-term follow-up, adjuvant chemotherapy, and radiotherapy are usually beneficial as high cases of recurrence have been reported.

List of Abbreviation

GCT: Granulosa cell tumors

Declarations

Ethical approval and consent to participate

Written informed consent for publication was obtained from the patient whose management is being reported.

Consent for publication

Both authors gave consent for publication of the work under the Creative Commons Attribution-Non-Commercial 4.0 license.

Availability of data and materials

All essential data supporting the findings of this case are available within the article. Additional data are available upon request from the corresponding author.

Competing interests

The authors declare no conflict of interest.

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Authors' contributions

All the authors were involved in the management of the patients and conceptualizing the report. OVC, JBB, NCD, MI, OAI and OOM wrote the first manuscript. OVC, AA, STO, and NJI, corrected the manuscript. All the authors agreed on the final manuscript. The manuscript has been read and approved by all the authors.

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