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Anita Madan
Associate Professor, Department of Gynaecology and Obstetrics, Govt. Medical College Amritsar, Punjab, India

Davinder Pal
Department of Gynaecology and Obstetrics, Govt. Medical College Amritsar, Punjab, India

Renu Bala
Senior resident, Department of Gynaecology and Obstetrics, Govt. Medical College Amritsar, Punjab, India

Prabhleen Kaur
Junior resident, Department of Gynaecology and Obstetrics, Govt. Medical College Amritsar, Punjab, India

Corresponding Author:
Prabhleen Kaur
Junior resident, Department of Gynaecology and Obstetrics, Govt. Medical College Amritsar, Punjab, India

Mucinous cystadenocarcinoma of ovary in pregnancy: A rare case report

Anita Madan, Davinder Pal, Renu Bala and Prabhleen Kaur

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Abstract
The ovary is a frequent site of neoplastic tumours that could be either benign or malignant. Ovarian cancer (OC) is one of the most commonly diagnosed cancers among women. Regretfully due to the broad spectrum of clinical behaviour and challenging diagnosis most cases are diagnosed at a late stage. On rare occasions, these tumors can grow to massive sizes if left untreated, worsening the prognosis of the patient. We hereby report the rare case of Primary Unilateral Mucinous Cystadenocarcinoma of ovary in a 23-year-old pregnant woman which was initially misdiagnosed as Hydatid disease. Albeit, it was a malignant tumour, it presented with torsion in the post abortal period with benign behaviour and normal levels of CA-125 which is usually elevated in epithelial ovarian malignancies. The mass was removed along with unilateral salpingoophorectomy and histopathologic study revealed a mucinous cystadenocarcinoma.

Keywords: Ovarian mucinous adenocarcinoma, ovarian cancer, metastasis

Introduction
Tumours of the ovary are common forms of neo plasia in women. Among cancers of the female genital tract, the incidence of ovarian cancer ranks next only to carcinoma of the cervix and endometrium [1]. The ovary is unique in range and variety of tumours that may arise from it [2]. Malignant tumours from other primary sites can also metastasize to it. Little is known about the trends or international variations in the occurrence of the various histological types of ovarian tumours. Studies are hampered by differences in diagnostic criteria used over a period of time from place to place [3, 4].
The second most common epithelial tumour of the ovary is the mucinous type. Mucinous cystadenoma accounts for 10-20% of all ovarian tumours. The recurrence of mucinous cystadenoma is very rare after complete excision.
Benign neoplasm of the ovaries are of epithelial origin in 50%, of all mucinous neoplasm 77-87% are classified as benign. They tend to be cystic in nature and 76% of mucinous tumours are multinodular while 24% are uninodular [5].
The risk of ovarian cancer is less clear than other genital cancers. Risk factors are nulliparity and family history [6].
It is rare before age 40, increases steeply thereafter and peaks at ages 65-75 [7]. The preservation of a normal ovary may be required to conserve fertility functions in a younger woman.
The incidence of ovarian tumor detected during pregnancy is 1/300 to 1/556 pregnancies. Of the ovarian tumors detected during pregnancy, the incidence of ovarian malignancy is 1/15,000 to 1/32,000 in the maximum reports [8]. Considering its rarity, we hereby report a case of pregnancy with Mucinous Cystadenocarcinoma of ovary in a 23-year-old female which was misdiagnosed as Hydatid cyst.

Case report
A 23 year old female, $G_3P_2L_2$ with 10 weeks 6 days period of gestation presented in OPD with chief complaints of amenorrhea since two months with progressively increasing abdominal mass disproportionate to the periods of gestation associated with heaviness in lower abdomen specifically on right side. Patient had a USG report documenting a large cyst with multiple daughter cyst of size 26×11 cm seen in abdomen suggestive of Hydatidcyst. She was being treated with anti-helminthics by the surgery department where the patient reported previously and was sent for gynaecological opinion.

The patient had pain over whole of abdomen, with no aggravating or relieving factors. There was no history of nausea, vomiting, fever, syncopal attack, bladder or bowel complaints. There was no history of discharge or bleeding per vaginum. Her previous menstrual cycles were normal. Her obstetric history was uneventful. All previous issues were alive and healthy. On general physical examination she was conscious and coherent. Her BP was 100/84 mm of Hg, pulse rate was 80/minute and she was afebrile. Abdominal examination revealed abdominal mass corresponding to 30 weeks of gestation.

Per vaginal examination: Uterus was 10 week size and a cystic mass was felt separate from the uterus arising from the pelvis filling the abdomen, mobile smooth in consistency felt in right lateral and anterior fornix. Left fornix was free. No nodular deposits were found.

USG documented anIntra-uterine pregnancy of 10 weeks with large cyst with multiple daughter cysts of size 26×11 cm in abdomen suggestive of hydatid cyst. She was managed on expectant line i/v/o pregnancy.

Two days later patient came to the gynae emergency with inevitable abortion. Evacuation and curettage was done on 22 June, 2019. She was advised CT abdomen at the time of discharge. Three weeks after abortion patient reported back with CT abdomen suggestive of large cystic multiseptatedabdmino pelvic mass with adjacent structural displacement? Ovarian in origin. Patient was admitted and evaluated further. MRI revealed huge septatedabdomino pelvic cystic mass probably right side. Tumor marker CA-125 was found to be 31.5 units/mL.

Patient was taken up for exploratory laparotomy. Peritoneal washings along with sub-diaphragmatic washings, washings from both left and right paracolic gutters and pouch of douglas were taken and sent for cytology.

Careful intraperitoneal exploration was done and biopsy of omentum was taken and sent for histopathology. A large cystic mass arising from right ovary with 2.5 times torsion of pedicle, about 40×30 cm, smooth in outline with bluish hue was seen. Capsule was intact. No adhesion, no prominent vessels were seen on the surface. No ascites was there. Right salpingophorectomy along with removal of cyst with intact capsule was done. Left ovary and tube were healthy. Biopsy from left ovarywas taken. Left ovary and tube were preserved as the patient was desirous of another pregnancy. Patient's post-operative period was uneventful and was discharged under satisfactory condition.

Histopathology report came out to be mucinous cystadenocarcinoma of ovary. H& E stain showed atypical cells arranged in back to back glandular and papillary with cystic dilated spaces containing mucinous secretions. Cells showed stratification, high N:C ratio, vacuolated cytoplasm, Vesicular chromatosis and prominent nucleoli.

Fig 1: Gross specimen showing oval cystic mass measuring 40×30 cm with smooth glistening greyish surface with bluish hue.

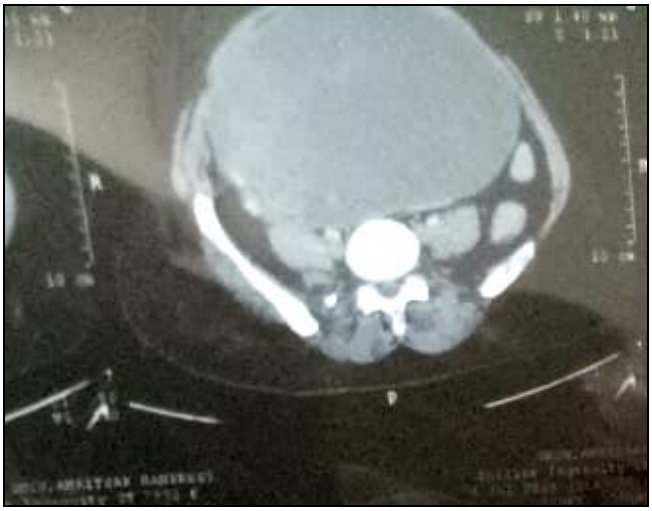

Fig 2: Computed tomography scan

Discussion

Mucinous tumours are the commonest large ovarian tumours having mean diameter of 16 to 19 cm. They grow into enormous size and are the largest gynaecological tumours [9]. They may macroscopically reach massive dimensions like in this patient it weighed 5kg and size 40 ×30 cm.

Mucinous tumors of ovary are the second most common type of surface epithelial tumor of ovary and account for approximately 15% of all ovarian tumors. They are divided into benign, borderline and invasive type. Approximately 80% of the mucinous tumors are benign, 10% are borderline and 10% are invasive carcinomas [10].

The incidence of malignant ovarian tumors in < 20 years age group is 22.6% in Indian scenario. [11]Present case is reported in 23 year old female.

A large retrospective series and a SEER database analysis have both shown that 79 % of mucinous tumors are unilateral. Our case also occurred as a unilateral tumour effecting right side of ovary [12].

Keeping in the mind the increase in number of cases reported as metastatic mucinous adenocarcinoma, it should be carefully differentiated from primary ovarian carcinoma. The incidence of primary mucinous adenocarcinoma is low. Features favouring primary ovarian carcinoma vs. metastasis are: unilateral, "expansile" pattern of invasion, complex papillary pattern, size > 10 cm, smooth external surface, microscopic cystic glands, necrotic luminal debris, mural nodules and accompanying teratoma, adenofibroma, endometriosis or Brenner tumor [13]. In present case, the mass was large and there was smooth external surface. There were no nodules on the surface. Usually bilateral masses, small size, capsular implant, surface involvement, nodularity, and hilar involvement favour metastasis.

Estimation of CA125 can help in identifying epithelial tumours of ovary. The normal value is 0 to 35 units/ml.CA 125 is found to be elevated in most of the cases reported in past but however interestingly our case did not show raised levels. Recent published literature revealed that the CA-125 is a blood test that is often, but not always, elevated with ovarian cancer. However, in younger women, CA-125 is extraordinarily inaccurate. It is not specific to ovarian cancer and may be elevated in other conditions. It should not be drawn just to see the level since it is not a reliable screening test for ovarian cancer. Inhibin and mullerian factor are other tumour markers which should also be added in the investigative protocol [14, 15]

The mucinous tumors of the ovary are usually early stage at

diagnosis (83%), in comparison to serous tumors (4%), and have a higher overall survival. However in advanced stages, the outcome of mucinous tumor is poor [16, 9]. Present reported case was Justin a 23 year old young woman. Hess *et al.* also suggested that advanced mucinous ovarian cancer had a worse outcome as compared to non-mucinous type, with advanced non-mucinous living 3 times longer than those with mucinous pathology [17].

Distant metastases are rare and survival is 95% for stage I and 32% for stages II or greater. In early-stage ovarian cancer, lymph node dissection is necessary to make a meticulous staging according to the International Federation of Gynecology and Obstetrics classification and to select an adequate adjuvant therapy. The effect of lymph node dissection on progression-free survival and overall survival in patients with advanced ovarian cancer is still unknown. The prognostic factors for stage I tumors are infiltrative invasion, high nuclear grade, tumor rupture [18].

Literature revealed that Mucinous neoplasms generally occur in young women and are diagnosed at an early stage, with 83% being diagnosed at stage I and only 17% at stage II or higher [16, 18]. Present reported case also showed consistent finding with reported literature as it was also diagnosed as Stage 1 a in younger age. The most common metastatic routes are via direct peritoneal seeding and lymphatic spread. Most stage I invasive mucinous carcinomas of the intestinal type with expansile growth pattern, like this patient, do not metastasize. Mucinous carcinomas of the infiltrative pattern of stromal invasion are more aggressive in comparison and account for the majority of metastasis [10].

The rate of recurrence following conservative and radical surgical procedures in low-stage and low-grade tumours are 9% and 11.6%, respectively; and disease-free and overall survival rates do not differ significantly. After surgery, the patient should be followed-up carefully as some tumours recur [19].

Management of ovarian tumours depends on the patient's age, the size of the cyst and its histo-pathological nature. Conservative surgery as ovarian cystectomy and salpingo-oophorectomy is adequate for benign lesions and for malignant lesions cytoreduction followed by exploratory laparotomy and then chemotherapy is necessary. Present case whichwas misdiagnosed as Hyadatid cyst by a previous Health care centre was treated by right salpingophorectomy as patient was strongly desirous of pregnancy.

Radiologically, it should be kept in mind that hydatid cysts can mimic many ovarian pathologies, particularly malignancies, and this should be considered during differential diagnosis [20].

Conclusion

Ovarian cancer remains the leading cause of gynaecologic cancer-related deaths. Every middle aged woman should go for screening for Gynecological malignancies. Pelvic Hydatid cyst should be considered in differential diagnosis of ovarian neoplasms.

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