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## Case Report

# A case report of serous cystadenofibroma in pregnancy

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### ABSTRACT

The presence of a giant adnexal mass in pregnancy is a very rare occurrence and is usually asymptomatic which is detected incidentally on a routine ultrasound examination of the first trimester or until it reaches a large size and becomes symptomatic. Serous cystadenofibroma is a cystic neoplasm containing fibrous component and ciliated epithelial cells surrounded by clear serous fluid with smooth or papillary surface with abundant vessels. We present a clinical case of a 24-year-old unbooked second gravida at 40 weeks 3 days who came in labour with a giant tumour of the right ovary. An emergency caesarean section was done followed by right salpingo-oophorectomy. Histopathological examination revealed the mass to be serous cystadenofibroma of the right ovary. We understood that proper antenatal evaluation and strict monitoring of tumour is necessary to decide the time and type of surgical treatment in order to avoid maternal or fetal complications.

**Keywords:** Serous cystadenofibroma, Adnexal mass, Pregnancy, Caesarean section

### INTRODUCTION

Adnexal masses are masses of ovary, fallopian tube or surrounding tissue. Adnexal masses during pregnancy are mostly functional usually resolving on their own before 3<sup>rd</sup> trimester.<sup>1</sup> If such cystic lesions persists till term then it indicates organic nature of the cyst, which are mostly benign.<sup>1</sup> Some non- adnexal masses which are present in a similar fashion include pedunculated fibroids, hydrosalpinx or para-ovarian cysts.<sup>2</sup> Some reported cases of giant ovarian cysts were wrongly diagnosed as ascites.<sup>3</sup> The histological type, cystadenofibroma, is rarely found, but is often serious, which can be further classified by WHO as serous, mucinous or mixed depending on the epithelium.<sup>4</sup> These cysts can be benign, borderline or malignant based on its epithelial proliferation and stromal components with size between 1-20 cm.<sup>4</sup> The rarity of the tumour encouraged us to report the case of an asymptomatic patient who presented at term in labour with

ovarian mass, which was removed during caesarean section.

### CASE REPORT

#### *Sociodemographic characteristics of the patient*

A 24-year-old unbooked Gravida 2 para 1 with previous normal vaginal delivery at 40 weeks 3 days reported to our emergency with lower abdominal pain since morning. She also gave history of right sided abdominal swelling for 7-8 months. The pain was continuous and progressive in nature. She did not have any complaints of per-vaginal leaking or bleeding.

She was a known case of hypothyroidism on medication. There was no significant surgical history. She had attained menarche at 13 years of age and previous menstrual cycles were normal. She had delivered a female child 4 years ago

via normal vaginal delivery which was uneventful. There was no significant family history.



**Figure 1: Abdominal distension of the patient along with term pregnancy at the time of admission.**

#### **Examination and diagnostic approach**

She was short stature with no pallor, icterus, clubbing, cyanosis or lymphadenopathy. There was no acne, facial hair or excessive body hair. Her routine systemic examination was normal.



**Figure 2: USG visualization of a large, cystic space occupying lesion done at the time of admission.**

On per abdominal examination, inspection showed a distended abdomen with linea nigra and striae gravidarum. Palpation revealed a huge abdominopelvic mass, cystic in nature with a dull note on percussion. Uterine size and fundal height could not be distinguished and estimated properly due to the mass. Similarly, presentation could not be estimated properly in per-abdominal examination due to the mass. Fetal heart sound was auscultated. On per-speculum examination, show was present. On per-vaginal examination, cervical os was 2-3 cm dilated, 50% effaced, presentation as cephalic with station vertex at -2 with bag

of membranes present. She only had one ultrasound report done from outside some hours earlier, which was suggestive of single live intrauterine fetus, at 37 weeks 3 days with cephalic presentation, posterior fundal placenta, adequate liquor and normal doppler.



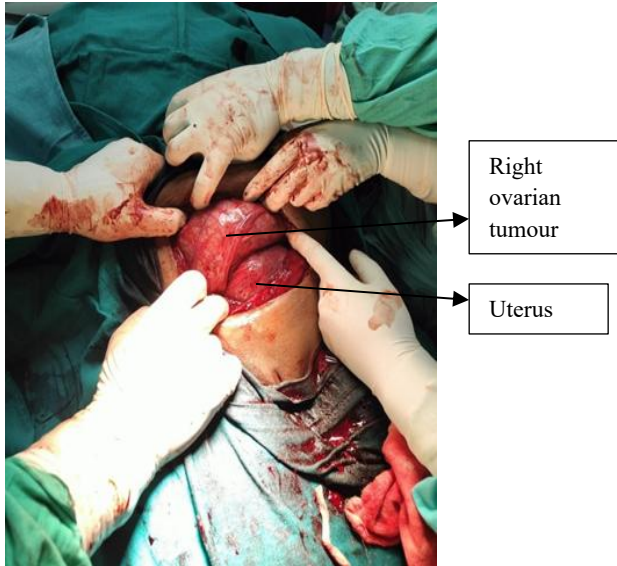
**Figure 3: USG showing simple, cysting lesion with low-level internal echoes done at the time of admission.**

It reported a cystic space occupying lesion measuring 18X17 cm in size, noted in lower abdomen extending up to right epigastrium suggestive of likely ovarian origin. No early gestational scan reports were available. A routine admission ultrasound was done by us which was suggestive of a cystic abdominopelvic space occupying lesion 20X19 cm in size. It was unilocular with no septations, papillary projections without being able to attach it to an organ, thereby indicating two diagnoses, either a giant ovarian cyst or a huge mesenteric cyst. There was no evidence of torsion on doppler. MRI is the radiological investigation of choice in such cases but in our case, the patient was in labour and hence, MRI could not be done.<sup>5</sup> However, mesenteric cysts are more common in the pediatric population and very rare in the adult population.<sup>1</sup> The fetal biometric parameters gave an estimated fetal weight of 2.6 kg with adequate liquor and normal doppler flow. We performed her blood investigations (shown in Table 1) and tumour marker tests (shown in Table 2).

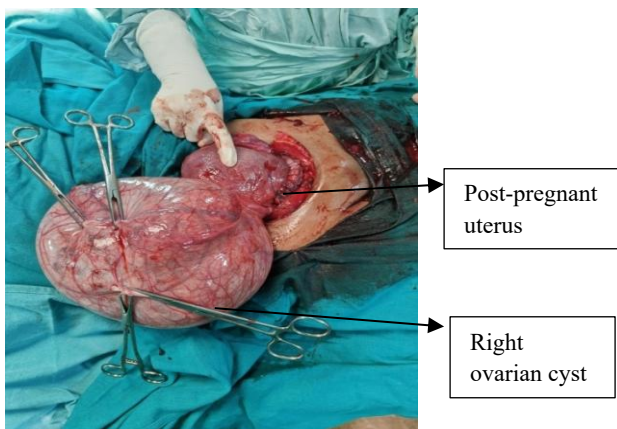
#### **Treatment**

After taking proper consent for oophorectomy and salpingectomy if needed, explaining the possibility of post-partum hemorrhage, need for blood transfusion, ICU admission and chances of infection requiring prolonged hospital stay, the patient was prepared for emergency caesarean section along with tubal ligation. Pfannenstiel incision was given. A single live baby was delivered by cephalic presentation, which cried immediately at birth with Apgar score 10 and 10 at 1 minute and 5 minutes respectively. Liquor was clear. Cord was clamped and cut. Placenta membranes were removed in toto by controlled cord traction. Uterus was closed in a single layer continuously with vicryl. No angle extension was observed. Bladder base was normal. The right ovary was

identified and evaluated, where the presence of a giant 25X25 cm fluid filled mass originating from it was noted. It completely took over the ovary along with the right fallopian tube and was merged with the mass because of which it could not be preserved.



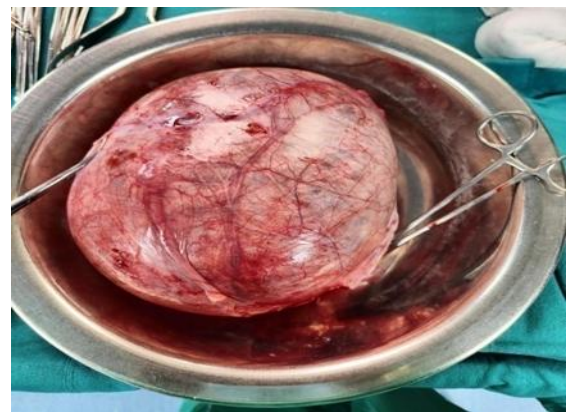
**Figure 4: Intra-operative picture of giant tumour of the right ovary seen partially during caesarean section.**



**Figure 5: Intra-operative picture of post-pregnant uterus in caesarean section with total exposure of the right ovarian tumour measuring approximately 25X25 cm.**

Hence, right salpingoophorectomy was done by serial clamping and cutting. This decision was taken as the cyst was large and incorporated the entire ovary leaving no spare healthy tissue for reconstruction, risk of bleeding after cystectomy and adhesences.

The right infundibulopelvic ligament was sutured with vicryl 1. Left sided tubal ligation was done by Modified Pomeroy's Method. The removal of the ovarian cyst was followed by exploration of the entire abdominal cavity. We could not find any evidence of ascites or any other abnormalities of the other intraperitoneal organs. The patient tolerated the procedure well and remained stable throughout the intraoperative period. Blood loss was average, and drain was placed in Pouch of Douglas. Post-operative urine was clear. The patient's abdomen turned flat immediately after surgery. The removed cyst measured 30 cm long axis in size, globular in shape with a smooth surface and soft consistency.



**Figure 6: Giant ovarian cyst after removal.**

**Post surgical vital signs**

Uterus involution was normal below the level of umbilicus, with normal lochia and no excessive per-vaginal bleeding. Drain was removed on Day 2 post-operatively. Routine analgesic and antibiotic coverage were given as per schedule. The post-operative period was uneventful and she was discharged on third post-operative day. She came for regular post-natal visits. Her histopathology report came to be serous cystadenofibroma of right ovary.

**Table 1: Laboratory parameters of the patient.**

Parameters	Previous records	On admission	Normal values
<b>Hemoglobin (g/dl)</b>	9.7	11.0	12-15.8
<b>Total count (µl)</b>	Not available	10,400	4,000-11,000
<b>Neutrophil count (%)</b>	Not available	83	40-70
<b>Platelet count (/µl)</b>	Not available	3.17 lac	1.5-4.5 lac
<b>PT (sec)</b>	Not available	17	12.7-15.7
<b>INR (sec)</b>	Not available	1.19	0.8-1.2
<b>Urine routine</b>		Normal	

Continued.

Parameters	Previous records	On admission	Normal values
TSH (mIU/l)	4.2	2.5	0.4-4.0
RBS (mg/dl)	102	92	<140
VDRL	Non-reactive	Non-reactive	
HIV	Non-reactive	Non-reactive	
HCV	Non-reactive	Non-reactive	
HBSAG	Non-reactive	Non-reactive	
Urea(mg/dl)	Not available	23.9	12.8-42.8
Creatinine(mg/dl)	Not available	0.47	0.6-1.2
Uric acid(mg/dl)	Not available	4.4	2.5-5.6
Sodium(mmol/l)	Not available	136	136-145
Potassium(mmol/l)	Not available	3.94	3.5-5.1
Blood group	Not available	A positive	

**Table 2: Tumour markers of the patient.**

Tumour markers	Result	Normal values
Alpha fetoprotein (AFP) (IU/ml)	38.08	0.0-8.5
Carcino-embryonic antigen (CEA) (ng/ml)	0.5	<=4.7ng/ml)
CA-125 (ng/ml)	25.34	0-35
Beta-HCG (mIU/ml)	218.4	
Lactate dehydrogenase (LDH)	244	115-221

## DISCUSSION

### Epidemiology

Adnexal masses in pregnancy account to about 0.2-2%.<sup>6</sup> The incidence of ovarian cysts in pregnancy is about 4.9%.<sup>1</sup> The most commonly occurring ovarian masses in pregnancy include corpus luteal cysts, theca lutein cysts, which resolves by 16 weeks.<sup>6</sup> Others include benign cystadenomas and mature cystic teratomas (dermoid cysts), rete ovarii tumors and endometriotic cysts in decreasing order of frequency.<sup>1</sup> Malignant tumors are relatively very rare among pregnant women.<sup>7</sup> Serous cystadenoma is the most common benign ovarian cyst and accounts for 60-75% of ovarian cysts.<sup>1</sup> The peak incidence of them occurring is between 20-40 years, usually due to unknown etiology.<sup>1</sup> They are not hormonally sensitive and do not resolve after 16 weeks of pregnancy.<sup>2</sup> Cysts having a size between 5-10cm and existing beyond 16 weeks require careful monitoring with USG, Colour Doppler and MRI. Serous cystadenofibroma is relatively rare which occurs in the age group between 23-80 years.<sup>4</sup>

In literature, some rare cases of ovarian mucinous cystadenoma with functional stroma have been reported, which produce hormones such as androgens, leading to virilization of pregnancy, fetal distress and premature birth.<sup>8</sup> Ovarian cysts are usually incidental findings in the first trimester, or until they attain a large size. They can then be removed along with an elective caesarean section in a single step or a two-step surgery most likely salpingo-

oophorectomy depending on the fertility status and the complication in the current pregnancy.<sup>8</sup> If the size of the cyst is more than 10 cm, surgical intervention is mandatory due to increased chances of torsion, malignancy and labour obstruction in pregnancy.<sup>6</sup> One of the worst complications occur, when the cyst undergoes torsion, which is common when it has a diameter ranging between 6-8 cm occurring between 10-17 weeks of gestation in about 60% of cases.<sup>1</sup> In about 38-60% of pregnant patients with torsion, the Doppler flow is normal on ultrasound.<sup>9</sup>

### Pathology

Most serous carcinomas originate from the fallopian tube and have an appearance similar to the glandular epithelial lining of the tube, while others like clear cell, endometrioid arise from endometriosis and resemble proliferative endometrium.<sup>10</sup> Serous cystadenofibroma is a benign epithelial adnexal tumour which is a rare entity and originates from the ovary and is less likely from the fallopian tube. They are mostly asymptomatic and are reported as an intra-operative incidental mass.<sup>5</sup> It is made up of both epithelial and fibrous stromal tissues which make up less than 2% of benign ovarian tumours.<sup>5</sup> It consists of cysts, glandular elements and fibrous stroma with absent or rare mitotic figures.<sup>5,4</sup> It is a surface epithelial tumour which can be further classified as serous, mucinous, endometrioid, clear cell and mixed. It can be benign, borderline or malignant, based on the degree of epithelial proliferation and stromal component.<sup>11</sup> Specific features of malignancy is seen on imaging in these tumours which include cystic, complex cystic or solid components separated by irregular, thick septae with papillary projections.<sup>5</sup>

Cystadenofibroma appear more severe in imaging than in reality due to their solid elements and thick septa in their structure.<sup>4</sup> On radiological imaging, it resembles a malignant neoplasm but the fibrous component gives a characteristic MRI appearance which differentiates it from malignant ovarian tumour.<sup>11</sup> The adnexal masses may have different characteristics. For instance, high risk masses tend to be solid, nodular with thick septa, while low risk ones are fluid filled anechoic cysts with thin walls.<sup>8</sup> Some tumours may appear as benign on imaging, but prove to be

malignant intra-operatively. Similarly, serous cystadenofibromas are benign ovarian tumours. They resemble malignant neoplasms on imaging.<sup>5</sup> They have characteristic features, which are concentric rings of calcification, called as Psammoma bodies.<sup>12</sup> Pregnancy also gives a confusing radiological picture in general, as the increased oestrogen and progesterone changes the USG appearance of tumors. For instance, it is difficult to differentiate endometriomas from borderline or invasive ovarian masses due to the presence of papillary projections in all of them.<sup>13</sup>

### **Diagnosis and imaging**

Transvaginal ultrasonography is useful to characterize the morphology of ovarian tumors.<sup>14</sup> However, transabdominal ultrasonography is preferred over transvaginal sonography during early weeks of gestation.<sup>15</sup> Numerous ultrasound mass scoring systems such as International Ovarian Tumour Analysis (IOTA) rules, ADNEX (Assessment of Different Neoplasia in Adnexa) model have been validated in pregnancy. In case of complicated anatomy during pregnancy, MRI can be used without a contrast.<sup>7</sup> Tumour markers can also be used for prediction of the type of tumour; however, their level fluctuates physiologically also in pregnancy and hence cannot be used for accurate diagnosis. CA-125, the epithelial cell tumour marker increases in first trimester, HE4 level decreases in pregnancy, except in third trimester. CA 19-9, a mucinous tumour marker increases slightly with gestational age. Inhibin A level increases in first trimester and in Down's syndrome. Inhibin B, AMH value increases in granulosa cell tumour and also physiologically in pregnancy. Beta-HCG value increases in pregnancy and also in germ cell tumours. Alpha fetoprotein (AFP), increases in pregnancy as well as in germ cell tumours, neural tube defects, and decreases in Down's syndrome. In case of dysgerminoma, LDH level increases, which also rises in severe preeclampsia and HELLP syndrome.<sup>2-8</sup> CA-125 is a tumor marker which is elevated in ovarian malignancy as well as in early pregnancy and early puerperium.<sup>7</sup>

### **Symptoms**

Patients with serous cystadenofibromas are mostly asymptomatic (25%) of cases, but occasionally lower abdominal distension, pressure or pain such as dyspareunia, bloating, constipation and vomiting may arise due to pressure effect.<sup>1-4</sup> These may mimic the physiological problems in pregnancy and hence could be in danger of being neglected by the patient.<sup>3</sup> Acute symptoms like pain may occur secondary to torsion, rupture or hemorrhage.<sup>12</sup> Large cysts can compress the gravid uterus, retard the intra-uterine fetal growth and can lead to premature delivery or abnormal presentation of the fetus.<sup>1</sup> Giant cysts can lead to postpartum haemorrhage.<sup>1</sup> They can also present similar to ascites due to their large size with flanks fullness, and demonstrable shifting dullness on abdominal examination.<sup>3</sup> A case was reported

of a 35 year old multigravida who presented at 38 weeks with per-vaginal leaking in first stage of labour. Her obstetric ultrasound was suggestive of polyhydramnios with ascites. Augmentation of labour was done, but there was poor progression and hence emergency caesarean section was done. Intra-operatively, there was no ascites, but a giant right sided ovarian cyst was seen extending up to epigastric region after exteriorizing the uterus. The cyst was decompressed and a right salpingo-oophorectomy was done. The histopathological study confirmed serous cystadenoma of ovary.<sup>3</sup>

### **Treatment**

The types of adnexal masses in pregnancy, which need surgical approach include dermoid cysts (32%), endometriomas (15%), functional cysts (12%), serous cystadenoma (11%), and mucinous cystadenomas (8%), and around 2% of them are malignant.<sup>8</sup> For cysts  $\geq 10$ cm, there is an increased risk of malignancy, torsion or labor obstruction in pregnancy. Hence, surgical removal is advisable. They should undergo laparoscopy or laparotomy avoiding intra-operative rupture spillage.<sup>12-16</sup> Ideally, laparoscopy is planned between 16-20 weeks of gestation to decrease the risk of miscarriage and preterm labour.<sup>9</sup> Laparoscopy is not recommended when the cyst is suspected of malignancy due to increased chances of spreading of malignant cells. Aspiration of the cystic contents should be avoided, due to increased risk of complications like infection, bleeding, rupture of the cyst or increased risk of peritoneal adhesion.<sup>1</sup> Laparoscopy is not advisable as gestational age advances due to the impact of uterine size on visualization.<sup>9</sup> There was a case report of a 24-year-old multiparous woman who presented at 20 weeks of pregnancy with abdominal distension. Anomaly scan revealed a large anechoic cyst. Tumour markers were sent. The patient was posted for diagnostic laparoscopy and laparoscopic cystectomy was done successfully at 20 weeks after explaining the remote possibility of miscarriage.<sup>6</sup>

In our case, laparotomy was preferred as it could be done along with the caesarean section. Moreover, we were unsure of the benign nature of the cyst pre-operatively. However, the best time for surgery is mostly the second trimester.<sup>6</sup> Ovarian cystectomy in an edematous ovary may be technically difficult and hence adnexectomy may be necessary, as was done in our case.<sup>16</sup> Frozen section biopsy is the gold standard for the diagnosis of cyst adenofibroma as it pre-empts aggressive surgery.<sup>5</sup> But, in our case, as the patient was in labour, she was taken up for emergency caesarean section which could not facilitate frozen section biopsy. There was a case report of a 27-year-old nulliparous woman with an ovarian cystic mass measuring 35X24 cm. Exploratory laparotomy with frozen section was done which gave an intra-operative picture similar to serous adenofibroma, but histopathology indicated stromal Leydig cell tumour and adenofibroma. She was also eager to conceive, hence IVF with oocyte donation was offered after complete ovarian suppression. She then came up

with a positive UPT test and an ultrasonography with 5 weeks of pregnancy.<sup>11</sup> In cases of a giant ovarian cyst complicating term pregnancy, an elective caesarean section can be done along with salpingo-oophorectomy, like in our case. There was a case report of a 28-year-old multigravida who presented at 27 weeks of gestation with acute onset of right sided abdominal pain. The USG was suggestive of right sided complex ovarian mass with intermittent Doppler blood flow, indicating ovarian torsion. She had a history of previous caesarean section with bilateral ovarian cystectomy which was done for right mucinous and left serous cystadenoma. She received steroid coverage for fetal lung maturity and right oophorectomy was performed, after which she received tocolytics and antibiotics. The histopathological diagnosis was of borderline mucinous tumour which was positive for CK20 and CDX2, suggestive of GI origin of the tumour. An elective caesarean section was planned for her at 37 weeks, which was followed by staging laparotomy and appendicectomy. Peritoneal washing and omental biopsy were negative for malignant cells. Post-natal MRI was indicative of no abnormalities in the GI system.<sup>17</sup>

A case was reported identical to our case in which a 34-year-old multiparous woman with previous two caesarean sections presented at 36 weeks with pain in abdomen and left ovarian cyst measuring approximately 10X15 cm. Emergency caesarean section was done along with excision of the giant tumour with left salpingo-oophorectomy.<sup>8</sup> Yet another case was reported in which a 32-year-old, booked multiparous woman with previous caesarean section, normal ultrasound reports and uneventful pregnancy, underwent elective caesarean section at 39 weeks of pregnancy. Intra-operatively, a small exophytic lesion was seen in the right ovary as an incidental finding. Excision was done and sent for examination. The histopathological study reported it to be cystadenofibroma with foci of endometriosis.<sup>4</sup> A retrospective study of all third trimester pregnancies with adnexal mass delivered via caesarean section along with cystectomy was done for outcomes which included any intra-operative complications as well as fetal outcomes. No reported intra-operative or post-operative complications and good fetal outcome were concluded in this study.<sup>18</sup>

## CONCLUSION

Ultrasound is definitely the most significant tool in the study of adnexal masses in pregnancy due to its safety and availability. However, definitive diagnosis can be made by histopathology. Ovarian cysts that coexist with pregnancy should be watchfully monitored; else a condition may arise when the patient may not know that she had tumour which had happened in our case.

Borderline ovarian tumours can be problematic for diagnosis pre-operatively, and the definitive confirmation can be done only after surgical excision followed by histological confirmation. A wait and see strategy is

applicable for asymptomatic ovarian cysts in pregnancy with benign features. In case of any masses with septa, solid components, papillae or nodules existing after 16 weeks, further investigations need to be done. Both open and laparoscopic approach can be adopted for ovarian masses in pregnancy depending on the tumour diameter, gestational age and surgical expertise. If a large adnexal mass is detected at the third trimester of pregnancy, it favours delivery via caesarean section to avoid the risk of shoulder dystocia and the advantage of one step approach of cystectomy/oophorectomy at the time of caesarean section. It does not increase complication. This also prevents further surgery after caesarean section in most of the patients. Hence to conclude, prenatal care, clinical evaluation as well as ultrasound examinations play an important role in identifying the complications during pregnancy. The tumor mass removal followed by histopathological evaluation is the gold standard to identify ovarian tumours and to reach an ultimate diagnosis to determine the appropriate conduct for each case. Moreover, there is no difference in fetal outcome as was evident in the current case of pregnancy with an adnexal mass.

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