



Decasualization of a haemorrhagic ovarian cyst during pregnancy mimicking malignancy

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Abstract

The incidence of ovarian tumour in pregnancy varies widely from 1 in 81 (1) to 1 in 8000 (2) live births in the earlier literature. On a more recent literature, an incidence of 1 in 505 (3) has been reported. Majority of the ovarian tumours found during pregnancy were benign but between 2.4 to 5.3% (4, 3) of those were found to be malignant. In recent times, detection of ovarian tumours during pregnancy was increased due to routine prenatal ultrasound checkups. As a result, there is an increased need to distinguish between a benign versus malignant ovarian tumour. This is often challenging as decidualised benign ovarian cyst may mimic features of malignancy. Other challenges lie in the decisions regarding the need to operation and when to operate on such ovarian tumours. Most symptomatic tumours or those showed borderline or malignancy features on ultrasound are ones likely to require surgery. Historically, pregnant women with symptomatic or suspicious masses underwent elective removal in the second trimester (12) and this is relatively safe. Any surgical Intervention beyond the 24 weeks's gestation is associated with poorer obstetric outcome such as spontaneous miscarriage, preterm labour or preterm premature rupture of membranes (13). The decision to operate hence is a challenging one especially when one needs to balance between the upstaging of the suspicious malignant ovarian mass and the wellbeing of the fetus. Here we report a case where decidualised haemorrhagic ovarian cyst without evidence of endometriosis showed excrecence which obliged us to perform surgery during pregnancy. As the findings were found in the 3rd trimester, the operation along with a caesarean section was carried out prematurely at 35 weeks of gestation to achieve fetal maturity, fetal wellbeing as well as the possibility of staging operation in the form of total abdominal hysterectomy oophorectomy if tumour was deemed malignant.

Keywords: ovarian tumour, pregnancy, Decidualization, malignancy

1. Introduction

The incidence of ovarian tumour in pregnancy varies widely from 1 in 81 ^[1] to 1 in 8000 ^[2] live births in the earlier literature. On a more recent literature, an incidence of 1 in 505 ^[3] has been reported. Majority of the ovarian tumours found during pregnancy were benign but between 2.4 to 5.3% ^[4, 3] of those were found to be malignant. In recent times, detection of ovarian tumours during pregnancy was increased due to routine prenatal ultrasound checkups. As a result, there is an increased need to distinguish between a benign versus malignant ovarian tumour. This is often challenging as decidualised benign ovarian cyst may mimic features of malignancy. Other challenges lie in the decisions regarding the need to operation and when to operate on such ovarian tumours. Most symptomatic tumours or those showed borderline or malignancy features on ultrasound are ones likely to require surgery. Historically, pregnant women with symptomatic or suspicious masses underwent elective removal in the second trimester ^[12] and this is relatively safe. Any surgical intervention beyond the 24 weeks's gestation is associated with poorer obstetric outcome such as spontaneous miscarriage, preterm labour or preterm premature rupture of membranes ^[13]. The decision to operate hence is a challenging one especially when one needs to balance between the upstaging of the suspicious malignant ovarian mass and the wellbeing of the fetus. Here we report a case where decidualised haemorrhagic ovarian cyst without evidence of endometriosis showed excrecence which obliged us to perform surgery during pregnancy. As the findings

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2. Case report

Ms WLS is a 36 years old lady in her first pregnancy with a family history of diabetes mellitus and no known history of endometriosis of her own. Antenatal booking bloods, 14th week Down's screening, fetal DNA test in the form of Safety 21 were entirely normal. As a result of her family history and age, oral glucose tolerance test performed at 28 weeks suggested the diagnosis of gestational diabetes. She remained a diet control gestational diabetic throughout her pregnancy with satisfactory control of her glucose and HbA1c throughout her pregnancy. During her first scan at 14 weeks, she was found to have a right adnexal elongated cystic mass 3.5x7.6cm with hypoechoic content and irregular thickening of the inner wall which measured 1.4x2.3cm, likely of right ovarian or tubal origin with no obvious suspicion of malignancy. A routine morphology scan by a private doctor showed the baby to be a female fetus with no gross abnormality but did not comment on the existing right adnexal mass. At 26 weeks, patient had a repeat scan in the hospital. A combination of transabdominal and transvaginal ultrasound scan showed the right adnexal cystic mass's size has increased to

6.8x5.2x5.9cm. The complex cystic mass was mainly of hypoechoic in nature with specs of solid / hyperechoic shadow within the cyst and a papillary growth projection of 3.6x1.9cm was seen within. (Figure 1 and 2)



Fig 1



Fig 2

Given the growth and nature of the cyst, suspicion of a borderline or malignant tumour explained to the patient. Ca125 was not performed explaining the non-specific nature of Ca125 during pregnancy. For further investigation for the suspicious ovarian cyst, a plain MRI abdomen was done at 28 weeks. Other than a singleton intrauterine pregnancy, it showed a 3.1x5.2x5.9cm (TSxAPxLS) predominant cystic mass (hypotense on T1 and hyperintense on T2-weighted sequence) at the right adnexa. The wall is slightly irregular with some nodularities. Image suggested a complex cystic lesion at the right adnexa with irregular thick wall with nodularity and suspicious eccentric signal change that may represent presence of haemorrhage or high proteinaceous content. Left ovary, spleen, adrenal, small and large bowel, bilateral kidney, liver and pancreas were all normal. There was no frank evidence of liver or peritoneal metastasis.

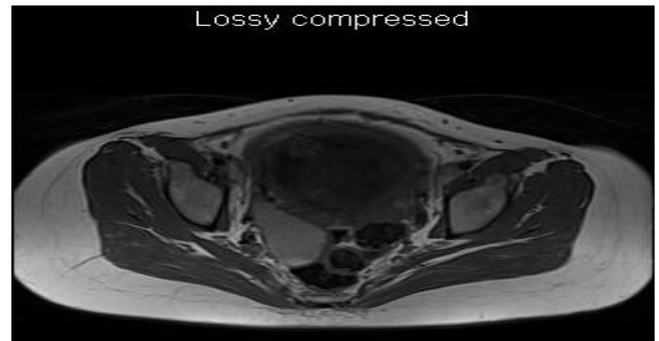


Fig 3: Axial T1 weighted image

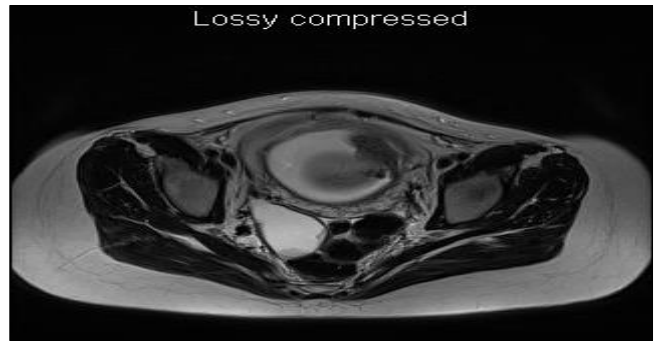


Fig 4: Axial T2 weighted image



Fig 5: Sagittal T2 weighted image

A repeat USG scan at 31, 32 and 34+6 weeks showed the complex lesion to be 5.2x7.5x4.5 cm (31 weeks), 6.26x5.93cm (32 weeks) and 5x6cm (34+6 weeks) respectively. Growth of the baby remains at 25 percentile throughout.

After a joint meeting involving obstetricians, gynaecological oncologists, patient and her relatives, decision was made to deliver the baby by laparotomy and Caesarean section with antenatal corticosteroid injection at 35 weeks on the balance of possible disease progression and fetal maturity. A unilateral ovarian cystectomy or salpingo-oophorectomy and subsequent frozen pathological section was also planned in view of further total abdominal hysterectomy, remaining salpingo-oophorectomy and staging to be carried out if the frozen section deemed borderline or malignant in nature. At 35 weeks, patient underwent a sub umbilical midline laparotomy. Lower segment caesarean section performed giving birth to a female baby who weighed 2.17kg and Apgar score of 4 and 7 at 1 and 5 minutes respectively. Subsequently right ovarian cystectomy was performed with cyst remain intact as the appearance of cyst suggested an endometriotic cyst. Further dissection of the cyst after its removal appeared to consist of a 6.5 unilocular cyst containing chocolate material with soft edematous nodules ranging from 2mm to 8mm lining in the inner surface of cyst wall. Immediate frozen section of the cyst to pathology confirmed the cyst to be of haemorrhagic cyst of the ovary which was surrounded by denuded stromal tissue with prominent decidual changes. As a result of this finding, no further surgical intervention was carried out and abdomen was closed in the usual fashion. Final pathology of the right ovarian cyst again confirmed the cyst was haemorrhagic ovarian tissue in nature with decidualisation with no evidence of endometriosis. There was no evidence of malignancy while there were also no signs of malignancy in the histopathology of the placenta. Patient and baby recovered well post operatively and discharged together on day 4 post procedure.

3. Discussion

To our knowledge, this is one of the few cases of decidualized benign ovarian haemorrhagic cyst mimicked malignancy to be reported in the literature. Decidualization is the conversion of the normal endometrium into a specialized uterine lining adequate for optimal accommodation of the gestation during pregnancy. This change is caused mainly by progesterone and involves hypertrophy of the endometrial stromal cells leading to thickening of the normal endometrium and giving rise to the decidua. Decidualized tissue can grow during pregnancy to acquire a gross appearance that macroscopically mimics a malignant tumor^[8]. Several reports have described decidualized ovarian endometriosis^[5, 11]. During pregnancy, decidualisation not only occurs within the uterine endometrium but also to extrauterine endometrium in particularly in areas of endometriosis. How decidualization occurs within a haemorrhagic cyst without the presence of endometriosis in our case remains uncertain, it is likely to be due to the decidualisation of ectopic sites, namely stromal ovary in this case which mimicked features of malignancy^[15]. Intracystic papillary excrescences which are vascularized on colour Doppler examination are a sonographic association with malignancy^[14] however they are also mimicked by decidualised cysts. These

findings during ultrasound examination creates a dilemma between further investigating a potentially benign mass, sometimes through invasive procedures that may complicate pregnancy, versus conservative observation of a potential malignant state. MRI has been suggested in providing more information and possibility to differentiate between a decidualised ovarian cyst from malignancy. However results from other studies found no characteristic pattern. In general, excrescence showed low intensity in T1 in general but may show intermediate or high intensity on T2 which is similar to our presented case. However, ovarian malignancies exhibit a variety of MRI findings regardless whether they are of the same histological type or of different histological type^[10] thus MRI maybe unable to definitely distinguish decidualization from malignancy. Other investigation such as Ca125 cancer antigen level is often used during investigation for ovarian tumour. It may be particularly high in malignant tumours, however during pregnancy, they are found to be normally elevated in most patients particularly in the third trimester hence they are not particularly useful in the diagnosis of ovarian malignancy^[17]. As distinguish between decidualization and malignancy remains difficult, the need for surgery often based on the size of the tumour, the symptoms that the patient may suffer, the level of suspicions the practitioner may have as well as the patient's general anxiety. Majority of the cysts including neoplasms are found in the early parts of the pregnancy and operations are usually performed in the second trimester, in particular 16-18 weeks. At this gestation, uterus is not as large to obscure view or increase operative difficulties hence laparoscopic approach is a possibility. It is also associated with less adverse events compare to those done above 23 weeks gestation which includes spontaneous miscarriage, preterm labour or preterm premature rupture of membranes^[13]. However decidualization of ovarian tissues only occurs during pregnancy. There is this possibility where vascularization in the papillary excrescences was not detectable in the first examination but present later in the pregnancy, while with malignancy, one would expect vascularization to be present from the first examination^[7]. Similar to our case, the ultrasonic presentation of papillary excrescences in ovarian cyst which mimic malignancy may appear beyond the ideal operative window. Once the diagnosis or strong suspicion of neoplastic tumour is made, its removal is imperative to prevent acute complications and to resolve the possibility of malignancy. However, surgical removal of ovarian tumours in the third trimester remains controversial. Manipulation of the uterus at this stage of pregnancy may lead to premature labour^[7]. Aspiration of cyst under ultrasound guidance for diagnosis and symptom relief while minimize risk of preterm labour have also been reported^[16], however there is a risk of fluid re-accumulation and the possibility of upstaging its malignancy staging if the tumour was malignant. In our case, the decision was made to deliver the baby via caesarean section at 35 weeks while remove parts of the cyst for frozen section and subsequent total abdominal hysterectomy and staging if frozen section was deemed malignant. At 35 weeks, it is globally accepted to have a good fetal outcome and prevented the chance of extreme prematurity while keeping the fetal wellbeing in mind, such decision limited the delay of diagnosis and management of suspected malignancy to the minimum.

4. Conclusion

Decidualisation of ovarian cysts during pregnancy is rare in pregnancy. It occurs to endometriotic cysts but even less common to ovarian cyst with no evidence of endometriosis. Its ultrasonic appearance often mimic those of malignancy but none of the investigations clearly differentiates one from another. Surgical intervention remains the key to diagnosis and in preventing cyst complication or resolve possible malignancy but timing of the operation remains crucial when they are found late in particular during the third trimester. Decidualized ovarian cysts should be added to the differential diagnosis when there is a suspicion of ovarian malignancy during pregnancy to reduce risk of early surgical intervention and subsequent risks of preterm labour and fetal extreme prematurity.

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