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Case Report

Delivering a female with missing cervix: a case report

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ABSTRACT

Authors received a case in our casualty, 26 years old, G2P1L1, with full term pregnancy, spontaneous conception, previous normal vaginal delivery, with labour pain. On evaluation, she was having uterine contraction, fetal heart rate normal, vitally stable, with no cervix on per speculum and per vaginal examination, with solid globular mass per rectum. She was shifted to emergency operation theatre with provisional diagnosis of uterine rupture or rectal perforation or pelvic mass. After delivering a live baby, uterus was intact, but there was an impacted mass in pouch of Douglas, it was a twisted ovarian mass, sent for histopathology examination. Post-operative period was uneventful, and patient was discharged with healthy baby with corrected pelvic anatomy.

Keywords: Absent cervix, Adnexal mass in pregnancy, Missing cervix, Ovarian malignancy in pregnancy, Pelvic mass in pregnancy

INTRODUCTION

Adnexal mass in pregnancy is an uncommon event, with an overall estimated incidence of 0.02 to 1%.^{1,2} Although rare, ovarian cancer is the second most common gynecological cancer in pregnancy with an incidence of 1: 12.500 to 1: 25,000 pregnancies.³ Most of these adnexal masses are diagnosed incidentally at the time of dating or first trimester screening ultrasound.4 During Csections adnexal masses are found in about 0.3% of the cases.^{5,6} Pain due to rupture, hemorrhage into the cyst, infection, venous congestion, or torsion may be of sudden onset or of a more chronic nature. Masses which grow in pregnancy tend to be climb upwardly out of the pelvic cavity, as the uterus increases in size. Occasionally, they may be incarcerated in the Douglas pouch throughout pregnancy, with subsequent compression of adjacent structures, namely, urinary tract and lower digestive tract, with subsequent symptoms from these organs. The incarceration of the mass within the pelvis can also be responsible for labor dystocia through obstruction the descent of the presenting fetal part. Most adnexal masses

in pregnancy are benign and mainly represented by functional cysts (follicular, corpus luteum, and theca lutein cysts), teratomas, and cystadenomas. Nevertheless, malignant adnexal tumors should also be considered. The risk of malignancy is estimated around 0-10%.^{5,7-9}

USG (ultrasonography) screening during the first trimester of pregnancy has founded many adnexal masses. If an adnexal mass is palpated during examination, USG is the preferred radiological method of confirmation because of its ability to differentiate morphology and categorize the mass. This will ultimately allow stratification of risk without compromising maternal or fetal safety. Doppler studies have been shown to be a useful for diagnosis of malignancy in ovarian tumor. The Doppler criteria show that malignant tumors will generally have lower blood flow impedance and higher blood flow velocity. These findings can also be seen in inflammatory lesions.¹⁰

Computerized tomography (CT) and magnetic resonance imaging (MRI) can be useful adjuncts when USG imaging is inconclusive. CT imaging provides better resolution for identifying non-obstetric causes of abdominal pain. Although CT imaging is relatively safe in pregnancy, it does expose the mother and fetus to 2-4 rads in a single examination.¹⁴

MRI is particularly useful for characterizing large masses, evaluating their relationship with adjacent structures and differentiating masses appearing to have adnexal location on ultrasound but actually having a different organ filiation.¹² MRI is also useful, when an advanced stage malignant tumor is suspected, requiring the assessment of the retro peritoneum, abdominal cavity, and lymph nodes. MRI appears to be safe in pregnancy, but caution is advised for its use in first trimester, as well as for the use of gadolinium contrast, because of the potential fetal risks.¹³

The level of CA125, a glycoprotein in serum, may also be elevated with other benign disease processes such as menses, uterine fibroid, and endometriomas. CA125 is typically elevated during the first trimester, but may be useful during later assessment or follow-up. A15 Other tumor markers helpful in stratifying germ cell malignancy, such as alpha-feto protein (AFP), Beta human chorionic gonadotropin (BHCG), and lactate dehydrogenase (LDH), are of limited value because they may be significantly altered by pregnancy alone. In general, tumor markers should be used with caution during pregnancy, particularly in the first trimester, because of the wide variation in results and interpretation.

The Royal college of obstetricians and gynecologists (RCOG) in one of its guidelines stated, "simple, unilateral, unilocular, ovarian cysts less than 5 cm in diameter have a low risk of malignancy. It is recommended that in the presence of a normal serum CA125, they can be managed conservatively." ¹⁷

Generally, there is disagreement among authors concerning the best management of adnexal masses in pregnancy, with some recommending observation and others favouring surgical management. Most ovarian masses identified in pregnancy will spontaneously resolve and aggressive surgical management is not required. Characteristic features favourable for resolution are: masses that are simple in nature by USG, less than 5 cm in diameter, and diagnosed before 16 weeks. 19,20 Larger masses or those with more complex morphology are less likely to spontaneously resolve and may represent a neoplastic process. Similarly, persistent adnexal masses into pregnancy are more likely to be malignant or may result in complications in pregnancy, like torsion, rupture, or obstruction of labor. 21

Surgical intervention can be delayed until the second trimester to avoid the period of greatest risk of druginduced teratogenicity in first trimester, spontaneous fetal demise, and acceptable operative field will be available, allowing minimal uterine manipulation and low risk of obstetric complications. A later surgery in pregnancy, at the end of second trimester or at third trimester, may be technically more difficult and result in an adverse obstetric outcome. When indicated for other reasons, caesarean section may also be an opportunity for the surgical management of adnexal masses.

When considering surgery for an adnexal mass in pregnancy, the surgeon must optimize both maternal outcome and fetal well-being while performing an expeditious removal of the mass. Pregnant women undergoing surgery are at overall risk of prematurity (up to 22%) compared with pregnant women not undergoing surgery, regardless of the route of the procedure.²² The main disadvantage of delaying surgery during pregnancy is the risk of the mass undergoing torsion, rupture, or infarction, acute abdomen, and most importantly the risk of malignant change in case of ovarian mass. If the mass turns out to be ovarian cancer, the treatment of the pregnant woman is similar to that of the non-pregnant women depending on the stage, gestational age, as well as staging and grade of the tumor.²³ In certain circumstances, it may be justified to remove the tumor only and await fetal maturation, while in some cases chemotherapy may even be given while awaiting pulmonary maturation.²⁴

CASE REPORT

A 26 years old patient, G2P1L1, with 9 months of amenorrhea with 36 weeks 4 days of gestation, spontaneous conception, previous normal vaginal delivery, unbooked, housewife, low socioeconomic status, came to casualty with chief complaint of labour pain since last few hours. There was no history of leaking or bleeding per vaginum. Fetal movements were adequate. She was not having any chief medical illness and no obvious surgery in past. On general examination, vitals were normal. On per abdominal examination, uterus was term size, with cephalic presentation, with floating head, with moderate to severe uterine contractions, with fetal hearts rates 126 beats/minutes, regular, beat to beat variability poor with no declaration. On per speculum examination, vagina was normal and healthy, but cervical tissue was not seen at all. Per vaginal examination done, but there was no cervical tissue felt. On per rectal examination, there was a hard, globular circular mass anterior to rectum, which was coming down with uterine contractions. Patient was planned for emergency laparotomy with provisional diagnosis of uterine rupture, pelvic mass, rectal mass etc.

Treatment

Foleys catheterization done, routine blood investigation sent, blood group cross match done. All possible diagnosis with their expected managements was explained to relatives. Midline vertical incision was given, there was no hemoperitoneum after opening

peritoneum, usual steps of caesarean section were performed and a live healthy male baby was delivered out, who cried on operation table. Uterine contour was maintained. Assistant was asked to re-examined, but finding were same as was in preoperative condition. Then, uterus was exteriorized; there was a culprit in pouch of Douglas in the form of impacted, twisted, huge right ovarian mass measuring. Mass was coming out with difficulty, it was clamped, cut and ligated. Mass was dark brown in color, $12\times8\times6$ cm in size, intact, smooth walled, solid-cystic in consistency. After external examination, this mass was sent for histopathological examination. Abdominal organs and lymph nodes were examined, no obvious abnormality was observed.



Figure 1: Intraoperative ovarian mass.



Figure 2: Exteriorized twisted ovarian mass.

Outcomes

Post-operative period was uneventful. Patient was reexamined on day 7, to reconfirm the adequacy of treatment. Patient was discharged with healthy baby with normal and corrected pelvic anatomy. Histopathology report was suggestive of benign endometrium. Patient was regularly followed in subsequent period.



Figure 3: Removed ovarian mass.

DISCUSSION

Adnexal mass diagnosed preconception or during first trimester period warrants conservative management in most of cases. In some cases, termination of pregnancy may require depending on size of mass and nature of mass, which is relatively easy in first trimester and early second trimester. But, as in this case, late second trimester and third trimester it becomes nightmare sometimes to manage such kind of rare cases in pregnancy.

Patient had a normal vaginal delivery in previous pregnancy and it was a spontaneous conception, so there and there was no history of surgery in past, then, there should be a normal cervical tissue in patient. Then, missing cervix was an acute event like in ruptured uterus or inversion of uterus, or chronic event like huge pelvic mass, which make a distorted pelvic anatomy.

Since earlier ultrasonography report could help us to make a direction for further management, but patient was unbooked and uninvestigated, made our path more difficult. Ultrasonography in third trimester for diagnosis of adnexal masses, have low sensitivity and specificity. So, USG was not much helpful at that time.

Since this case patient was in active labour with good uterine contraction with floating head with missing cervix, normal vaginal delivery was not possible, so decision for emergency laparotomy was taken.

As compare to Pfannenstiel incision, midline vertical incision was given. This could help in exploration of abdominal cavity in case of malignant mass.

In this case, mass was benign in nature and no other abnormality was found in abdominal cavity, no other intervention was done and patient was followed up as in other cases. Patient was absolutely fine after 1 year of follow-up, which was satisfactory outcome.

CONCLUSION

Intrapartum abnormal position of cervix raises suspicious of abnormal pathology in patient. Pain in abdomen (non-obstetrical cause) should raise suspicious of non-obstetrical pathology in patients. Most adnexal masses, which appear in pregnancy, are functional and asymptomatic and resolve spontaneously. In asymptomatic pregnant women without signs of complications, conservative management is the most suitable option. When strong clinical suspicious of malignancy or presence of acute abdomen or severe clinical manifestation occur, surgical treatment is indicated.

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