Luteoma of Pregnancy - Mimicking a Malignant Ovarian Mass

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Summary

Luteoma of pregnancy (LP) are uncommon, benign, tumor like condition of the ovary, believed to be developed as the consequences of pregnancy induced hormonal changes and spontaneously regress postpartum. They are asymptomatic and incidental finding during surgery or imaging. They can pose a challenge for diagnosis and management plan because most of them present as malignant tumors of ovary. To avoid unnecessary surgery during pregnancy, if there is clinically high index of suspicion of tumor being pregnancy luteoma, can manage it conservatively during antepartum period and radiologically follow-up these suspicious masses during postpartum. Management of LP can vary from case to case and depends mainly on clinical situation. They usually present in the second and third trimesters of pregnancy. They are usually solid masses and frequently bilateral. Some patients may have elevated serum testosterone levels and display features of virilization in mother and female fetus. However, in some cases which present atypically or acutely due to the complications like rupture, torsion or haemorrhage into the mass lesion which requires immediate surgical exploration for diagnosis as well as timely management.

We present a 24-year-old woman, primigravida with 27 weeks of gestation, who was referred to the hospital with sudden distension of abdomen, abdominal discomfort and vomiting since 1month. Sonography and MRI showed large cystic lesion arising from left ovary, gross ascites and mild pleural effusion. The patient underwent surgery to remove this mass considering the imaging findings were suspicious for neoplasia. Histology and immunohistochemistry revealed pregnancy luteoma.

Keywords: Luteoma of pregnancy (LP), Mass mimicking ovarian malignancy, Ovarian tumors during pregnancy, Antepartum exploratory laparotomy, Virilizing tumors of pregnancy.

Introduction

The American College of Obstetrics and Gynecologists (ACOG) has released guidelines that describe the diagnostic approach and management of adnexal masses occurring outside of pregnancy. However, guidelines that dictate physician's approaches to females with incidental adnexal masses during pregnancy remain vague. Having to consider both the pregnant and fetus when making decisions regarding the management plan makes it more complicated. The main concerns with pregnant female who develop adnexal masses are - pregnancy complications and malignancies; timely management in this case is essential, without jeopardizing the health of the fetus. According to a recent study, adnexal masses are discovered in 1 per 76–1 per 2328 deliveries.

Luteoma of pregnancy (LP) was first described by Sternberg and Barclay in 1966. In general, luteomas are asymptomatic, and they are incidentally found during imaging and peripartum surgeries like cesarean section or tubal sterilization. Luteoma of pregnancy is a benign, hyperplastic tumor-like condition of the ovary. It may be unilateral or bilateral, 1/3rd is bilateral.[1] Hyper secretion of androgens occurs in about 25% of women with pregnancy luteoma, among them 10% to 50% of the patients may display some clinical findings associated with hyperandrogenism and when the masculinized mothers gave birth to female babies they showed features of virilization (approximately 60% to 70%).[2] LP can pose a challenge for diagnosis and management plan because most of them present as malignant tumors of ovary.[3]

Case Report

Hereby presenting a case report of 24 years old, primigravida with 27 weeks of gestation, who was referred to the hospital with sudden distension of abdomen, abdominal discomfort and vomiting since 1 month. Patient had a past history of vaginal reconstructive surgery- Z plasty for transverse vaginal septum at the age of 13 years, which got failed and again she underwent surgery for the same after marriage, following which she conceived spontaneously. On per abdomen examination ascites was present, uterus 26 weeks size, relaxed, FHS heard. Mass felt at left flank extending towards left hypochondriac area. Ultrasonography(USG) shows a single live intrauterine gestation of 27 weeks and 10.2x11.4x10.9 cm heterogeneous echo texture lesion at left lumbar region extends into left iliac region. Few cystic areas with thin septation and solid component shows vascularity. Moderate ascites and mild right sided pleural effusion was noted. Magnetic resonance imaging (MRI) of pelvis shows 16x12x14.8 cm heterogeneous signal intensity lesion with internal necrotic / haemorrhagic areas in left lumbar region up to inferior pole of spleen and splenic flexure. Moderate to gross ascites was present. Tumor markers were - CA 125 (1259 U/ml), AFP (129.1 ng/ml),

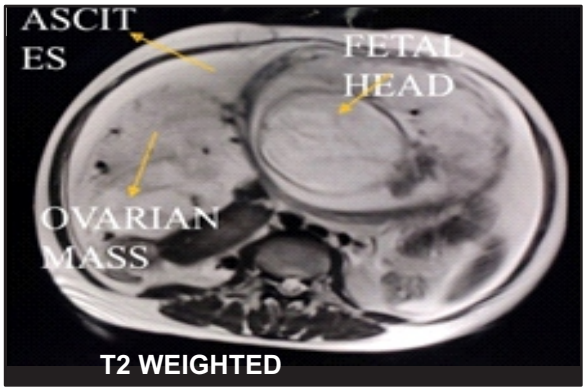

Figure 1: T2 weighted image showing left ovarian mass in relation to gravid uterus

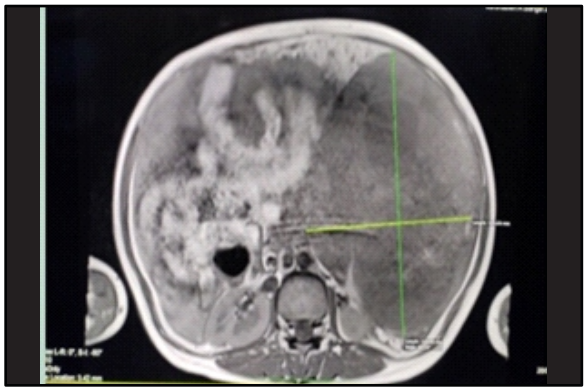

Figure 2: T2 weighted MRI showing left ovarian 16x12x14.8 cm solid cystic, well encapsulated mass with moderate to gross ascites. Right ovary is normal.

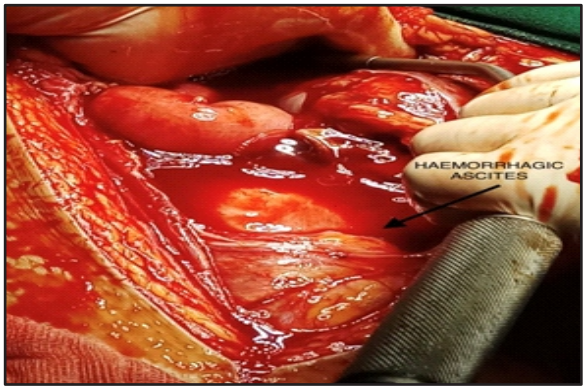

Figure 3: Intra operative image showing hemoperitoneum

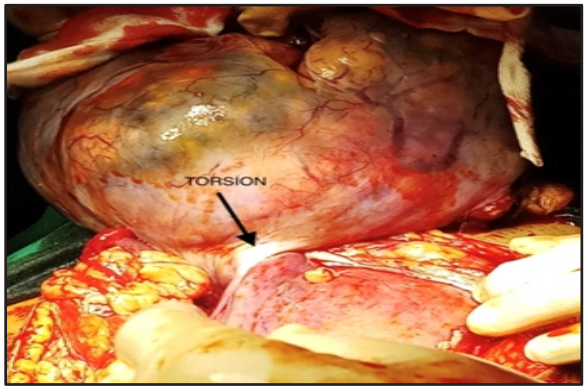

Figure 4: Intra operative image showing torsion of left ovary

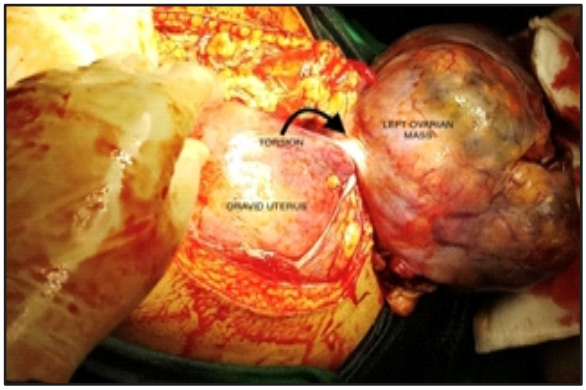

Figure 5: Intra operative image showing ovarian mass in relation with gravid uterus

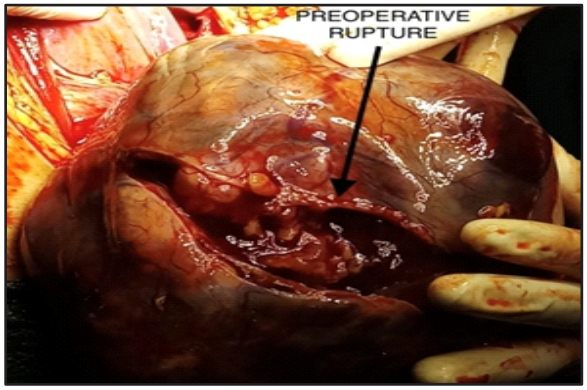

Figure 6: Intra operative image showing preoperative rupture of the ovarian mass

bhCG(16367 IU/ml), CEA(0.66 ng/ml), He4 (62.72pM). ROMA (Risk of Malignancy Algorithm) was 15.4%-high risk. Ascitic fluid cytology was suggestive of reactive mesothelial cells and negative for malignancy. Patient had hemoglobin 7.6 gm/dl and received two packed cells preoperatively.

Since there was radiologically, serologically and clinically high index of suspicion of malignancy, the decision was taken for surgery. On exploratory laparotomy 500 ml haemorrhagic ascites was drained out. Uterus was 26 weeks sized, relaxed with visible fetal movements. Approximately 25x15 cm mass, which had 360 degrees of torsion and preoperative rupture, was seen arising from the left ovary. Mass was removed along with adherent omentum and sent for frozen section. Rest of the abdomen was normal on gentle exploration.

Frozen section report was suggestive of luteoma of pregnancy. Postoperatively FHS monitoring was done and patient was managed with adequate analgesia, tocolytics(isoxsuprine) and progesterone (Duphaston). Fetal well being assessed by USG on 2nd post operative day.

Final histopathology and immunohistochemistry confirmed the diagnosis of LP. Patient was discharged on 6th post operative day

with good fetal and maternal condition. She delivered a full term baby of 2.8 kilograms by elective caesarean section. There were no signs of virilization in the newborn.

Discussion

LP is a rare condition, which resemble malignant tumors of ovary. They are believed to be a result of luteinized stromal cell hyperplasia. Approximately 50% of the times it is multinodular and in one third of the patients it is bilateral.

Luteoma of pregnancy have variable size at presentation, ranges from microscopic to over 20 cm in maximum diameter in literature and our patient had luteoma of 25 cm. Luteomas are most often clinically indolent and discovered incidentally. However, can rarely present as acute abdomen due to ovarian torsion, rupture or hemorrhage into the mass. They may be hormonally active and secrete androgen, which is responsible for virilization of both mother and female fetus. Rodriguez et al reported a case with gross ascites and elevated CA125 level in pregnancy.[4] Massive ascites and an elevated CA125 level in these type of cases resemble malignant tumor. These phenomena have been rarely reported in pregnancy luteoma cases. The increased expression of CA125 may be induced by mechanical stimulation/irritation of the mesothelium.[4] The solid ovarian tumor could physically irritate the peritoneum and stimulate the overproduction of peritoneal fluid. Rubinstein et al., suggested that the ascites results from a discrepancy between the arterial supply to a large tumor mass tissue and the venous and lymphatic drainage of the same mass, leading to stromal edema and transudation.[5] Tan ML et al reported torsion of tumor leading to its rupture and intra-peritoneal bleeding which required blood transfusion and immediate surgical exploration.[6] Even our patient presented similarly. Thus, decision regarding appropriateness of surgical management should be made by the clinician.

In a review of 11 cases of adnexal masses in pregnancy, most common tumor being mature cystic teratoma, found in 40 patients. Others are serous cystadenoma/cystadenofibroma (11 cases), mucinous cystadenoma (16 cases), and corpus luteal cyst (3 cases). Malignant or potentially malignant tumors constitute 6.1% of tumors, including a serous cystadenocarcinoma, serous borderline tumors (19 cases), an immature teratoma, a Sertoli-Leydig cell tumor and a Juvenile granulosa cell tumor. In general the malignant adnexal masses encountered during pregnancy is 3–6%.

Management of LPs varies and it depends mainly on clinical presentation, the character and the size of the tumor, period of gestation at presentation and choice of the patient.[7] Non-obstetric surgery during pregnancy posts additional concerns to anaesthesiologists. The chief goals are to preserve maternal safety, maintain the pregnant state and achieve the best possible foetal outcome. The choice of anaesthetic technique and the selection of appropriate anaesthetic drugs should be guided by indication for surgery, nature and site of the surgical procedure. Anaesthesiologist must consider the effects of the disease process itself and inhibit uterine contractions and avoid preterm labour and delivery. Foetal safety requires avoidance of potentially dangerous drugs and assurance of continuation of adequate uteroplacental perfusion. Until date, no anaesthetic drug has been shown to be clearly dangerous to the human foetus. The decision on proceeding with surgery should be made by multidisciplinary team involving anaesthesiologists, obstetricians, surgeons and perinatologists. Indications for surgery are clinical and radiologic suspicion of malignancy and patient presenting as acute abdomen due to ovarian torsion, rupture, haemorrhage into the mass. If the doubt arises regarding the diagnosis of pregnancy luteoma, then surgical exploration to be performed antenatally or post delivery and most frequent procedure being a unilateral salpingo-oophorectomy. Whenever the surgery is done intra operative frozen section for histological assessment is compulsory. This is because in case frozen section report is inconclusive and if diagnosis of malignancy is questionable, we can proceed with conservative management rather than more aggressive radical surgery. If final histopathology suggestive of malignancy, surgical staging should be performed postpartum.[8] Our patient underwent exploration because clinical and radiological features were suspicious of malignancy, patient had sudden abdominal distension due to ascites, abdominal discomfort and raised tumor markers.

Whenever the clinical judgment and radiological features are in favour of a luteoma, our management option should be conservative during antepartum and follow-up with imaging during postpartum, as there is spontaneous resolution after delivery.

Conclusion

Luteomas due to its clinical resemblance with malignant neoplasms, complicates its diagnosis and treatment. With high index of clinical and radiological suspicion for LP, conservative management during antepartum period and follow-up with imaging during postpartum period is an acceptable management strategy which will avoid unnecessary surgical intervention leading to pregnancy complications. However, in some instances with atypical and acute presentations, surgical exploration may be necessary to rule out malignancy and to provide timely treatment.

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