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Case Report

Beyond the ectopic – pregnancy luteoma as a rare case of acute abdomen in pregnancy

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ABSTRACT

Luteoma of pregnancy is a rare benign tumor attributed to the nodular hyperplasia of theca interna cells in the ovaries. Although the exact pathophysiology is still unknown, it is mainly implicated to the elevated levels of beta human chorionic gonadotrophin in pregnancy and pre-existing endocrinopathy. Here we are presenting the case of a female with an unlocalized pregnancy who presented in the emergency with symptoms of acute pain abdomen and presence of free fluid in the abdomen and was taken up for laparotomy in view of acute abdomen and possibility of ruptured ectopic pregnancy but the intraoperative picture to our surprise was entirely different. Intraoperatively, large solid cystic mass of 6 cm was found and salpingo-oophorectomy was done which was later confirmed as pregnancy luteoma on histopathology.

Keywords: Luteoma, Pregnancy, Ectopic

INTRODUCTION

Pregnancy luteoma is a benign ovarian tumor. It can present as an incidental finding, an ovarian mass, hyper-androgenism or acute abdomen. It is an extremely rare condition and its occurrence has been underestimated due to its asymptomatic nature with most patients going unnoticed. It is a self-limiting condition and mostly regresses after pregnancy termination.[1] Thus, it needs to be differentiated from other adnexal masses in a pregnant female for appropriate management as well as to avoid over-treatment.

CASE REPORT

A female in her early 20's, second gravida, para one presented to the gynaecology emergency with complaints of moderate to severe pain in the left lower abdomen and a positive urine pregnancy test. Her period of gestation was four weeks and five days, had conceived spontaneously and presented with a rising level of serum beta human chorionic gonadotrophin (beta hCG). There was no history of polycystic ovarian disease and previous menstrual cycles were regular. At presentation, her vitals were stable. On per abdominal examination, there was mild- moderate distention and tenderness in the left iliac region. Per speculum examination revealed no bleeding or discharge and per vaginal examination suggested a closed cervical os with cervical motion tenderness and left adnexal tenderness. There was no pallor clinically and her routine blood workup including renal and liver function tests as well as coagulogram was normal with a haemoglobin level of 12g/dl. She had two beta hCG values done at a gap of 48 hours which were 376 and 833 mIU/ml respectively, raising more suspicion. Ultrasound was suggestive of a left adnexal mass of about 5×6 cm size and presence of free fluid in the abdomen. The patient did not agree for a culdocentesis and opted for surgery. Hence, a decision for

laparotomy was taken owing to her clinical presentation and the possibility of a ruptured ectopic pregnancy. Intraoperatively, 1.2 litres straw coloured ascitic fluid was drained which was sent for cytology. The left adnexa had a large solid to cystic irregular mass adherent to the ovary, measuring 6×7 cm with a few haemorrhagic areas with no salvageable ovarian tissue, raising suspicion of an ovarian malignancy or ovarian ectopic pregnancy (Figure 1). There was no evidence of torsion. The uterus and right ovary were normal looking.

Thus, staging laparotomy was performed. Left salpingo-oophorectomy was done and omental and peritoneal biopsies were taken. On cut section, it was an irregular solid-cystic lesion along with areas of haemorrhage with few papillary projections, features highly suggestive of a malignant mass. Her post-operative period was uneventful and all tumor markers were sent to assess for any possibility of malignancy and to obtain the baseline values for follow up if needed, but they all except beta hCG came out to be insignificant. On post operative day 5, an ultrasound was done and findings were suggestive of an intrauterine gestational sac with no fetal pole. no other abnormality was detected on the ultrasound. Repeat beta hCG levels were 1697 mIU/ml. On postoperative day 6, patient had spontaneous bleeding per vaginum followed by complete abortion which was confirmed by an ultrasound. Patient was subsequently discharged. On follow up, ascitic fluid cytology came out to be negative for malignancy and the histopathology report was suggestive of a luteoma of pregnancy. The patient was kept on regular follow up and was fine thereafter for 3 years after which she was lost to follow up.

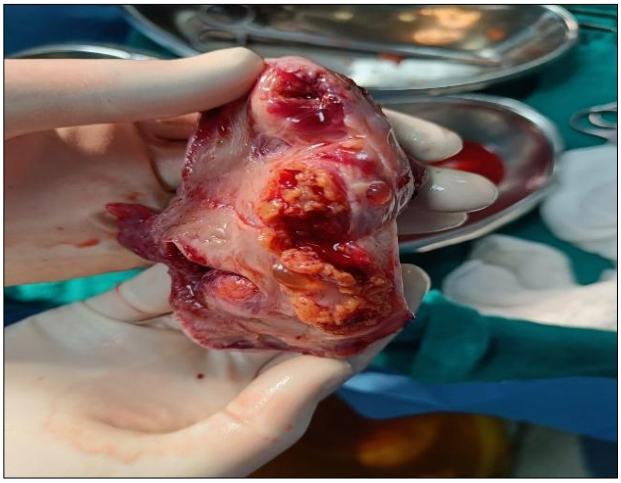

Figure 1: Intraoperative image of the left adnexal mass.

DISCUSSION

The incidence of adnexal masses in pregnant females is around 2-20 per 1000. Most of them are benign masses, including luteoma of pregnancy. It mostly occurs in third and fourth decade of life and predisposing factors include previous history of luteoma, multiple pregnancies, advancing maternal age, polycystic ovarian syndrome, etc. Although the most common ovarian tumor associated with pregnancy is dermoid cyst but other benign conditions like simple cysts, functional haemorrhagic cysts and rarely, hyper stimulated ovaries, luteoma of pregnancy, hyperreactio lutenalis are also encountered.[1,2] Luteoma of pregnancy usually regress spontaneously after 2-3 weeks of delivery. It is mostly asymptomatic, also known as sub-clinical luteomas, in which case they can either go unnoticed or present as a constant mass on regular antenatal ultrasounds, and may spontaneously regress after delivery but can also present as acute abdomen in cases like torsion or tumor rupture or with signs and symptoms of hyperandrogenism. Hyperandrogenic states in first trimester are mainly due to pre-existing hormone releasing polycystic ovarian syndrome whereas luteoma and hyperreactio-lutenalis present in second trimester.[1,3]

Macroscopically, it presents as a solid brown to yellowish fleshy tumour with haemorrhagic areas. Their size can vary from microscopic to 20 cm large masses. They are bilateral in around 30-50% cases. Microscopically, it consists of sheets of cells, polygonal in shape, arranged diffusely and containing abundant eosinophilic cytoplasm which form follicles containing colloid-like material. Its pathophysiology is still unclear but it is thought to arise from excess proliferation of luteinized cells under the influence of beta hCG.[3]

About 25-30% of luteomas are androgen secreting but only 10-50% of them manifest clinically. It may also cause virilisation of the female fetus in around 60% cases while the remaining 40% escape its effects owing to the protective effect of placental aromatase.[3] The excess hormone production from the tumour may lead to features of hyperandrogenism in the mother including deepening of voice, acne, hirsutism and clitoromegaly along with perinatal complications including high risk of miscarriage in first trimester and an increased risk of premature delivery in third trimester. Postpartum complications such as suppression of lactation may also be seen.[4]

Due to the solid nature of this tumor, various differentials like granulosa cell tumor, Sertoli-leydig cell tumor, thecomas and various solid ovarian tumors should be kept in mind as it is difficult to differentiate these tumors from pregnancy luteomas on the basis of imaging.[2]

The management of luteoma of pregnancy is case dependant. In symptomatic patients presenting with unresolving virilisation symptoms, pressure symptoms or an acute abdomen, operative management is required. In asymptomatic cases, a conservative approach can be considered provided adequate follow up is possible. Patients should follow up with repeat ultrasound and falling androgen levels at about 3-4 weeks post-delivery as pregnancy luteoma usually regresses spontaneously after delivery.[1-3] On the other hand, for a patient presenting with acute abdomen, other possibilities have to be ruled out, the

commonest being ruptured ectopic pregnancy, acute appendicitis, ovarian torsion, luteoma torsion, ruptured cyst, and malignancies. Ultrasound findings and culdocentesis may help compare the various differentials although any patient with deteriorating vitals and falling haemoglobin with acute abdomen requires laparotomy. Tumour markers should be evaluated if there is any suspicion of malignancy. Luteoma of pregnancy, being a rare condition, is often a diagnosis of exclusion and medical professionals should be aware about the tumour as a cause of acute abdomen in pregnancy to be able to diagnose it and avoid overtreatment. We as gynaecologists should thus be on the lookout for similar possibilities to avoid unnecessary laparotomy and hence reduce patient morbidity.

CONCLUSION

Luteoma of pregnancy is a rare benign tumor attributed to the nodular hyperplasia of theca interna cells in the ovaries. Although the exact pathophysiology is still unknown, it is mainly implicated to the elevated levels of beta human chorionic gonadotrophin in pregnancy and pre-existing endocrinopathy like polycystic ovarian disease and hyperthecosis. Though it is a benign mass and resolves on its own, it may present as acute abdomen and encounter with rare conditions like this pose a diagnostic pitfall and make it important for the clinicians to be on the lookout for such possibilities to avoid potential serious complications.

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