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Case Report

Hyperreactio luteinalis: benign disorder masquerading as an ovarian malignancy

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ABSTRACT

Hyperreactio luteinalis (HL) refers to pregnancy related moderate to marked enlargement of the ovaries due to multiple benign theca lutein cysts. It is caused due to elevated Human chorionic gonadotropins leading to maternal complications such as preeclampsia and preterm delivery may result. We report case of a 24 years old lady, G3P1A1L1 with spontaneous twin pregnancy at 13 weeks + 4 days gestation presented with chief complaint of lower abdominal pain on exertion for 5 days. Ultrasonography (USG) showed a large left ovarian mass in Pouch of Douglas pushing uterus up and extending into the left side of midline upto costal cartilage. It showed multiple thick septations with vascularity pointing towards malignancy. CA-125 was elevated to 193U/ml. Laparotomy was undertaken. Intraoperatively, bilateral huge, congested, bosselated, multicystic ovarian masses were present which replaced normal ovaries and appeared malignant. Bilateral oophorectomy was done. Specimens received for histopathological examination comprised of two large multilobulated, dark brown, ovarian masses with intact glistening capsule. Serial sections through both the masses showed thin walled, multiloculated cysts with smooth inner lining, filled with thin clear to hemorrhagic fluid. On microscopic examination diagnosis of Hyperreactio luteinalis, bilateral ovarian masses were made. HL can be misinterpreted on USG or laparotomy as ovarian malignancy resulting in unnecessary surgical intervention.

Keywords: Enlarged ovaries, Hyperreactio luteinalis, Theca lutein cyst, Spontaneous twin pregnancy

INTRODUCTION

Hyperreactio luteinalis (HL) refers to pregnancy related moderate to marked enlargement of the ovaries due to multiple benign theca lutein cysts.

It is caused due to elevated Beta human chorionic gonadotropins (hCG) or pituitary gonadotrophins. It is associated with gestational trophoblastic diseases, multiple pregnancy, use of ovulatory induction drugs and fetal hydrops.[1,2]

CASE REPORT

This is a case of 24-year-old lady G3P1A1L1 with spontaneous twin pregnancy at 13 weeks +4 days gestation. She presented with chief complaint of lower abdominal pain on exertion for 5 days. Per abdomen examination revealed tenderness. Ultrasonography (USG) showed large multicystic mass of size 28x15cm arising from left ovary in pouch of Douglas pushing uterus up and extending into the right side of midline upto costal cartilage. It showed multiple thick septations with

vascularity pointing towards malignancy. Both ovaries were not made out separately. Minimal free fluid was present in pelvic cavity. Beta hCG levels were markedly elevated. CA-125 levels were elevated to 193U/ml. Patient underwent laparotomy and intraoperatively, bilateral huge, congested, bosselated, multicystic ovarian masses which replaced normal ovaries were seen which appeared malignant. Bilateral oophorectomy was done. Specimens received for histopathological examination comprised of two large multilobulated, dark brown, ovarian masses with intact glistening capsule. Larger mass measured 16.5x8.5x6.5cm with weight of 580gm. Smaller ovarian mass measured 15x7x5.5cm with weight of 280gm (Figure 1).

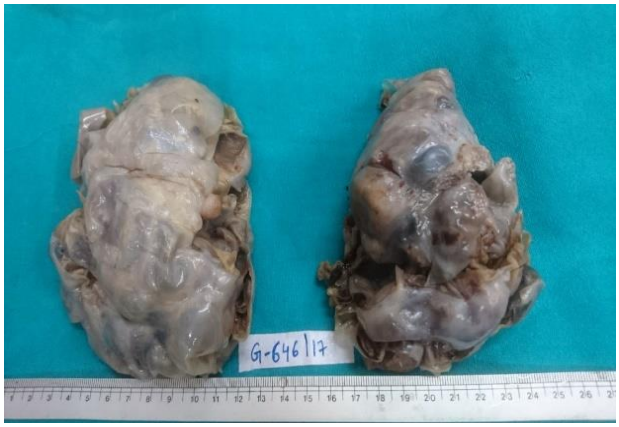

Figure 1: Gross examination: bilateral ovarian masses showing well encapsulated, glistening nodular external surface and cut section showing multiple variable sized thin walled multiloculated cysts.

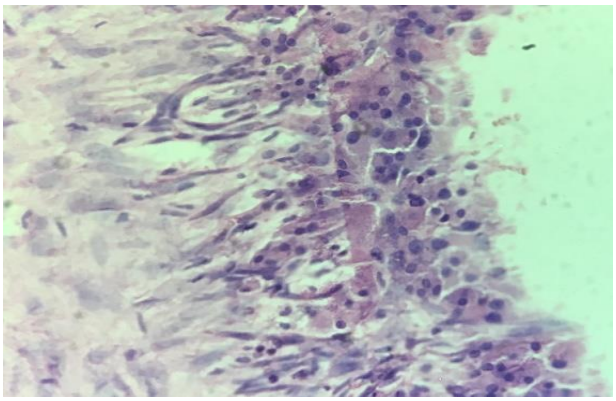

Figure 2: Microscopic examination: both the ovaries showed multilayering at places with occasional cysts showing flattened attenuated lining.

Serial sections through both the masses showed thin walled, multiloculated cysts with smooth inner lining, filled with thin clear to hemorrhagic fluid. No nodularities or papillary excrescences were seen. Microscopically, both the ovarian masses showed multilayering by theca lutein and granulosa cells with occasional cysts showing flattened attenuated lining. Ovarian stroma was edematous (Figure 2).

DISCUSSION

Hyperreactio luteinalis (HL) is self-limiting condition, follows benign course and presents with bilateral enlarged multicystic ovaries. It is seen usually in spontaneous multiple gestation, but few cases are also seen with singleton pregnancy[1-3] HL can present in any trimester but invariably regresses during postpartum period.[1,4]

The pathogenesis of HL is due to exaggerated response of ovaries to hCG which is produced by cytotrophoblast. Levels of hCG are maximum during 9th week so HL is seen in the 1st trimester of pregnancy.[2] Risk factors which cause elevated hCG levels are ovulation induction drugs, multiple gestation, gestational trophoblastic disease (molar pregnancy) and fetal abnormalities viz hydrops. We present a case of HL with marked elevated hCG levels with torsion ovaries in 2nd trimester. Our patient had a classical history of spontaneous twin pregnancy conceived without ovulation induction drugs, early second trimester gestation, markedly elevated hCG levels and bilateral pelvic masses inseparable from ovaries. However, difference from classical case was that both ultrasonography and intraoperative findings were more in favor of malignancy.

Various clinical findings in HL include hyperemesis gravidarum, hyperthyroidism, hirsutism, maternal virilization (due to hyperandrogenism), eclampsia, hemoperitoneum.[1,5,6] HELPP syndrome (hemolysis, elevated liver enzymes and low platelet count) syndrome develops subsequently in complicated HL.[7] It can variably be asymptomatic or give rise to significant complications like ascites, torsion and rupture with intra-abdominal bleeding.[1] Patient in our case report presented with lower abdominal pain only as a symptom. Ultrasound and intraoperative findings were pointing towards malignancy. Our case was complicated by torsion leading to hemorrhage and edema in the ovarian parenchyma.

Morphologically differential diagnosis includes ovarian tumors especially mucinous cystadenoma, ovarian hyperstimulation syndrome (OHSS) associated with preexisting polycystic ovaries and acute Meig's syndrome.[6-9] HL consist of many thin walled lutein cysts with large solid area which needs to be differentiated from mucinous cystadenoma which have less rounded locules with less solid component.[9,10] OHSS typically develops in early first trimester patients usually in those with ovulation induction which is often associated with ascites whereas HL develops later in pregnancy when levels of beta hCG are at maximum levels with no associated ascites.

Some cases remain asymptomatic in entire course of pregnancy and may be missed in cases of vaginal delivery. So, as a routine during caesarean intraoperative inspection of adnexa should be done to rule out ovarian

pathology. HL responds best to conservative management if it remains uncomplicated. When complications are detected during pregnancy, cyst aspiration may be done under ultrasonic guidance.[6] No surgery is indicated in HL unless it is important to remove infarcted tissue, control hemorrhage or diminish androgen production in virilized patients.[9] In our case bilateral oophorectomy was done due to hemorrhage and torsion of bilateral ovarian masses and suspicion of malignancy.

CONCLUSION

Diagnosis of HL is important to identify and manage maternal complications such as preeclampsia and preterm delivery. HL can be misinterpreted on USG or laparotomy as ovarian malignancy resulting in unnecessary surgical intervention. HL can produce symptoms any time during the pregnancy or may be seen incidentally during cesarean section if it escapes torsion and rupture. Hence it is necessary to be conversant with this entity due to benign nature of these grossly enlarged multicystic masses masquerading as malignancy.

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