

# Hemorrhagic Infarction of Hyperreactio Luteinalis in Spontaneously Conceived Singleton Pregnancy: An Uncommon Cause of Late Third-Trimester Ovarian Accident

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## Abstract

Ovarian masses complicate about 1%–2% of all pregnancies while posing diagnostic and therapeutic challenges. Most are benign tumors but occasionally, functional cysts, such as theca lutein cysts, may be encountered in pregnancy. These are bilateral ovarian masses associated with high levels of  $\beta$ -human chorionic gonadotropin, thus seen mostly in the settings of trophoblastic diseases, multiple gestations, or following treatment for subfertility. It has also been reported in spontaneous singleton pregnancies. Clinical presentation is variable, ranging from asymptomatic conditions discovered incidentally to acute gynecological emergencies due to ovarian accidents. We report the case of a 29-year-old gravida 2, para 1 + 0, with estimated gestational age of 39 weeks plus 1 day, who had a previous emergency lower segment cesarean section (ELSCS) 3 years ago and presented with 1-h history of sudden-onset, sharp, right-sided, abdominal pain of increasing intensity. There were no history of trauma and no evidence of labor. On examination, she was a young gravid woman, at term, in severe painful distress. Baseline investigations were done, and she had ELSCS and exploratory laparotomy with a right salpingo-oophorectomy for twisted and hemorrhagic right adnexa discovered. Pathologic evaluation of the right ovary revealed an enlarged, dark brown, lobulated ovarian mass comprising multiple cortical cysts separated by a thin wall and containing dark brown fluid, with gray brown and hemorrhagic solid areas. A histologic diagnosis of hemorrhagic infarction on the background of multiple theca lutein cysts in pregnancy was made. She had a good postoperative recovery.

**Keywords:** Hyperreactio luteinalis, ovarian accident, term pregnancy, torsion

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## INTRODUCTION

Ovarian masses presenting in pregnancy may pose diagnostic and therapeutic challenges. Most are benign tumors such as cystadenomas and mature teratomas. Occasionally, malignant tumors are encountered. Rarely, functional cysts such as theca lutein cyst may occur in pregnancy, also called hyperreactio luteinalis.<sup>[1]</sup>

Hyperreactio luteinalis are bilateral enlarging ovarian masses, associated with high levels of  $\beta$ -human chorionic gonadotropin ( $\beta$ -hCG), thus seen mostly in the settings of trophoblastic diseases, multiple gestations, or following treatment for subfertility. It has also been reported in association with spontaneous singleton pregnancies.<sup>[2,3]</sup>

The clinical presentation is variable, ranging from asymptomatic condition discovered incidentally to acute gynecological

emergency due to ovarian accidents secondary to torsion, hemorrhage, or rupture, necessitating exploration laparotomy irrespective of the gestational age of the pregnancy.<sup>[4]</sup>

## CASE REPORT

A 29-year-old gravida 2, para 1 + 0, with estimated gestational age of 39 weeks + 1 day, who had an emergency lower segment cesarean section (ELSCS) 3 years ago on account of fetal distress, malposition, cephalo-pelvic disproportion,

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and postdate, presented to the labor ward with a 1-h history of right-sided abdominal pain that was sudden in onset, sharp, severe, and radiating to the thigh. There was associated increasing frequency and intensity. There was also no known aggravating or relieving factor nor pain in any other part of the body. She had no history of trauma to the abdomen or abdominal manipulations prior to the onset of symptoms. There was no history of passage of show, drainage of liquor, fever, vaginal bleeding, or other genitourinary symptoms, and fetal movement was regular.

The pregnancy was desired, was spontaneously conceived, and was diagnosed with a positive pregnancy test, confirmed with an obstetric scan done at 8 weeks of gestation. The pregnancy was booked for antenatal care at 20 weeks of gestation. She had two more ultrasound scans which were normal and confirmatory of viable singleton gestation. She was planned for vaginal birth after cesarean section.

She had no other remarkable gynecologic or other past medical history and no history of blood transfusion in the past.

Physical examinations revealed a young woman in severe painful distress, not pale, anicteric, with no peripheral cyanosis, not febrile, and no pedal edema. Her pulse rate was 96 beats/min, full volume, regular, and synchronous with other peripheral pulses, and blood pressure was 130/80 mmHg in supine position. The respiratory rate was 20 cycles/min. The chest was clinically clear.

The abdomen was enlarged and moved with respiration, but there was a transverse suprapubic scar. There was marked generalized abdominal tenderness. The symphysio-fundal height was 39 cm, consistent with an intrauterine pregnancy of 39 weeks and compatible with her gestational age by date.



**Figure 1:** Intraoperative finding of a huge reddish brown right uterine adnexa

The fetal lie was longitudinal with cephalic presentation, and the whole of the fetal head was palpable per abdomen. No contractions were palpated, and regular fetal heart tone was heard. The vulva and vagina were normal. The cervix was not effaced, and the cervical os was closed.

A preoperative clinical assessment of acute abdomen in a parous young woman with one previous cesarean section likely due to scar dehiscence was made to rule out ovarian accident at term. Baseline investigations including pelvic scan, full blood count, serum electrolytes, urea, and creatinine were done and found to be all within normal ranges. Blood was grouped and cross-matched in preparation for surgery. She was counseled and an informed consent was obtained from her for an exploratory laparotomy and ELSCS.

The intraoperative findings were a live male neonate weighing 3.85 g with Apgar scores of 8 and 9 at the 1<sup>st</sup> and 5<sup>th</sup> min, respectively. A gangrenous right ovary and Fallopian tube measuring 15 cm × 15 cm was found and surgically removed [Figure 1]. The left ovary was enlarged, measuring 8 cm × 8 cm, whereas the left Fallopian tube was unremarkable.

The right salpingo-oophorectomy specimen was fixed in formalin and sent for histopathological analysis. She had a satisfactory postoperative recovery and was discharged 5 days later for follow-up.

Gross pathologic evaluation revealed a dark brown, enlarged, lobulated ovarian mass, measuring 11 cm × 7 cm × 5 cm with 6 cm-long Fallopian tube attached to it, weighing 162 g. Cut sections revealed multiple variably sized cystic cavities mainly located in the cortex, with the largest measuring up to 3 cm in diameter separated by a thin wall and containing dark brown fluid with gray brown solid hemorrhagic areas [Figure 2].

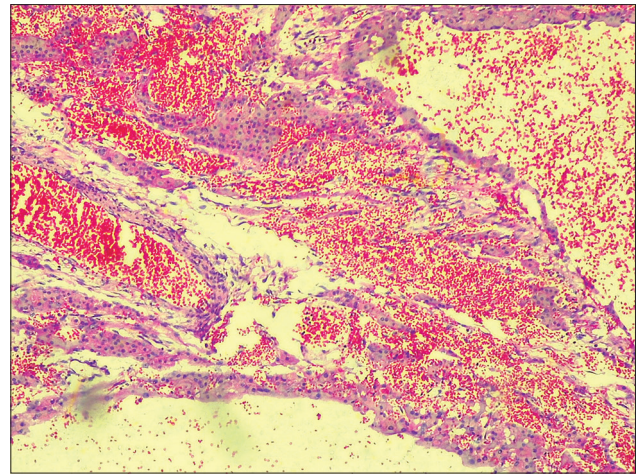
Hematoxylin and eosin sections showed variably sized cystic follicles lined by stratified-to-attenuated luteinized granulosa cells, beneath which are luteinized theca cells. Other



**Figure 2:** Cut sections of the ovarian mass showing multiple cortical cystic follicles with thin walls and gray brown and hemorrhagic areas



**Figure 3:** Photomicrographs showing multiple follicles lined by luteinized granulosa cells with focal hyperplasia and hemorrhage in the wall (H and E, ×40)



**Figure 4:** H and E section showing cystic follicles lined by luteinized granulosa cells, luteinized theca interna cells, and extensive areas of hemorrhage and necrosis (×100)

areas showed extensive hemorrhage, vascular congestion, and coagulative necrosis [Figures 3 and 4]. Sections of the Fallopian tube showed similar areas of hemorrhage, congestion, and necrosis. A diagnosis of hemorrhagic infarction on the background of multiple theca lutein cyst in pregnancy was made.

## DISCUSSION

Ovarian masses complicate 1%–2% of all pregnancies. Majority of those are benign conditions such as cystadenomas and mature teratomas, with malignant tumors being extremely rare.<sup>[1]</sup> Occasionally, multiple theca lutein cysts are encountered; these may mimic malignant tumors clinically and radiologically, often necessitating undesired surgical exploration.<sup>[5]</sup>

Multiple theca lutein cysts are characterized by bilateral cystic ovarian enlargement and hyperplasia of granulosa cells lining the cyst and luteinization of the granulosa and theca interna cells. This complication is seen commonly in association with trophoblastic diseases, multiple gestations, or in pregnancies following treatment for subfertility. Although uncommon, it has also been reported in spontaneously conceived singleton pregnancies.<sup>[6]</sup>

It is associated with elevated level of, or increased sensitivity to  $\beta$ -hCG, but other factors probably play a role.<sup>[7]</sup> About 25% of cases are associated with maternal virilization due to increased production of ovarian androgens by theca cells. As in the majority of cases, the present case was free of virilization.<sup>[8]</sup>

Some cases of multiple theca lutein cysts in pregnancy may present with symptoms related to hypertensive disorders of pregnancy, HELPP syndrome, and hyperthyroidism.<sup>[9,10]</sup> There was, however, no symptoms or signs of such conditions in this case. The patient has remained asymptomatic till the pregnancy was carried to term. Most cases remain asymptomatic throughout pregnancy and are only recognized at cesarean

delivery for unrelated indications, or may spontaneously regress few weeks postpartum.<sup>[3,11]</sup>

The index case was complicated by torsion prior to presentation. Like other ovarian masses, hyperreactio luteinalis is associated with the risk of undergoing torsion; this happens when it is twisted around its vascular axis, thereby compressing and blocking the venous and lymphatic drainages, leading to vascular congestion, ischemia, and hemorrhagic infarction/necrosis of the ovary.

Those presenting with acute abdomen secondary to ovarian accidents such as torsion, hemorrhage, and rupture constitute surgical emergencies and require immediate exploration laparotomy irrespective of the gestational age at presentation.<sup>[12]</sup> Most cases of multiple theca lutein cysts that escaped major complications usually regress spontaneously few weeks postpartum, thus surgical management should be as conservative as possible.<sup>[13]</sup> However, there is some risk of recurrence in subsequent pregnancies.<sup>[4]</sup> The left ovary of the index case showed marked regression, measuring 3.4 cm × 2.5 cm × 2.3 cm, just 2 weeks following the delivery as shown by the pelvic ultrasonography conducted during the postnatal visit.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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