Case Report

Polyhydramnios, an Unusual Complication in Pregnancy with Hyperreactio Luteinalis: A Case Report

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Reports of hyperreactio luteinalis in a spontaneously conceived pregnancy are uncommon. Hyperreactio luteinalis complicated by polyhydramnios without an identifiable cause has never been reported. The authors report a case of a 25-year-old, gravida 4, para 2-0-1-2, 16-week pregnant woman who presented with bilateral ovarian enlargement. The beta-human chorionic gonadotropin hormone was at a normal level. An exploratory laparotomy with biopsy was performed at 17 weeks of gestation. Polyhydramnios without an identifiable cause was detected at 30 weeks of gestation. A full-term healthy baby was delivered. Intrapartum and postpartum were unremarkable and both ovaries spontaneously regressed.

Keywords: *Hyperreactio luteinalis, Polyhydramnios*

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Hyperreactio luteinalis (HL) is a rare condition in pregnancy characterized by bilateral moderate to marked cystic ovarian enlargement. The association of ascites fluid and maternal virilization has infrequently occurred[1]. A true incidence of HL in a normal singleton pregnancy is not known. HL is usually associated with a high beta-human chorionic gonadotropin (β-hCG) level production or abnormal sensitivity of the β-hCG receptor[2]. Therefore, HL cases have been reported with multiple pregnancies, trophoblastic disease, after assisted reproductive treatments for infertility and association with rhesus incompatibility[2-4]. HL is usually not reported in singleton pregnancies and is generally diagnosed in the third trimester or found incidentally during a cesarean section operation[4-6].

Amniotic fluid is an important structure for fetal development. Its function can be categorized into three groups. First, its physical function serves to prevent fetal injury and regulate temperature. Second, the functional function is to regulate fetal skeletal movement, swallowing, and respiration. The functional function prevents the fetus from pulmonary hypoplasia and joint contracture. Last, the amniotic fluid maintains fetal homeostasis that includes maintaining fetal membrane integrity, inhibiting uterine contraction, preventing infection, and maintaining cervical length and consistency[7]. The amount of amniotic fluid is controlled by the dynamic equilibrium between the production of fluid from fetal lungs and the urinary tract, the flowing in and out during fetal swallowing and the absorption of the fetal membranes[8]. Polyhydramnios is a condition that presents with an abnormally increased amount of amniotic fluid. The incidence of polyhydramnios in the general population of pregnant women is around 0.2 to 1.6%[9]. Antenatal polyhydramnios can be diagnosed by clinical impression and confirmed by ultrasonographic measurement. Usually the amniotic fluid index (AFI) is the method to measure the fluid volume using ultrasonography. Polyhydramnios is defined as an AFI of more than 25 cm. The causes of polyhydramnios can be explained by excessive production and/or reduced utilization. The degree of polyhydramnios can vary from mild to severe. The perinatal mortality rate is directly dependent on the degree and cause of polyhydramnios in each case[10].

Although HL in pregnant women has been reported, most cases are associated with complications such as diabetes and hypertension[3,11-14]. An association of HL in normal pregnancy and polyhydramnios without an identifiable cause has never been reported. The authors experienced a case of HL with an unknown cause of polyhydramnios during a singleton pregnancy where an operation was performed during pregnancy and a healthy child was delivered.

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Case Report

A 25-year-old, gravida 4, para 2-0-1-2, pregnant Thai woman was referred to our hospital for bilateral ovarian tumor at 16 weeks gestational age. Enlargement of bilateral ovaries was recognized during her routine antenatal care. She had a spontaneously conceived and normal singleton pregnancy. Her history was unremarkable and she had not used medication for ovulation induction. Laboratory data showed normal results for the complete blood count, urinalysis, renal, liver and thyroid function tests. No proteinuria was detected. The serum β-hCG level was 59,681 mIU/mL, which was compatible with the normal range for her gestational age. Transabdominal ultrasonography showed a bilateral multicystic ovarian tumor without any solid part. The left ovary measured 8.2x9.1x7.8 cm and the right ovary measured 11.6x11.9x7.0 cm. No free fluid was detected in the pelvic cavity (Fig. 1). A detailed fetal anomaly scan showed a singleton male fetus with an appropriate weight for gestation and normal anatomy. The patient was scheduled for surgery because of the large size of the masses and the masses were suspected to be mucinous tumor. An exploratory laparotomy using a midline incision was performed at 17 weeks of gestation and revealed bilaterally enlarged multiple ovarian cysts without ascites and no suspected metastatic lesion (Fig. 2). A wedge biopsy was performed at the left ovarian cyst wall to rule out malignancy. A frozen section revealed probable luteoma.

After the operation, her clinical condition was stable and the patient was discharged from the hospital on the seventh postoperative day. Permanent microscopic sections demonstrated a granulose cell tumor. The patient routinely followed antenatal care at a referral general hospital and was sent to the hospital again at 30 weeks of gestation because her doctor suspected polyhydramnios. Ultrasonography revealed an appropriate weight for the gestational age of the fetus with a normal detailed anomaly scan and polyhydramnios (AFI = 35 cm). The 100 g oral glucose challenge test result was within normal limits. She was informed to attend ultrasonographic and routine antenatal care follow-up. No further complications were detected. An elective cesarean section was performed at 38 weeks of gestation and she delivered a healthy male infant without signs of virilization weighing 3200 g with Apgar scores of 9 and 9. Total amount of amniotic fluid was about 2,000 ml. During the cesarean section, the ovaries were slightly enlarged with multicystic appearance (Fig. 3). The placenta was grossly normal. At two and six months postpartum, the pelvic ultrasonographic study was negative. Both ovaries were unremarkable. The patient was healthy at the 1-year follow-up visit with a normal menstrual period.

Conclusion

HL commonly presents as bilateral ovarian enlargement, which is identified by ultrasonography

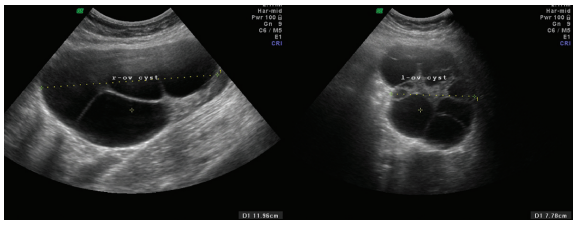

Fig. 1 Left and right ovaries show multiloculated ovarian tumor without any solid part.

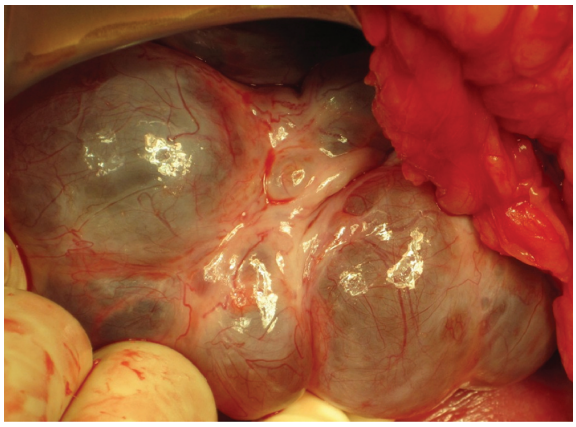

Fig. 2 Operative finding of left ovary at 17 weeks of gestation shows multiple ovarian cysts without ascites and no suspected metastatic lesion.

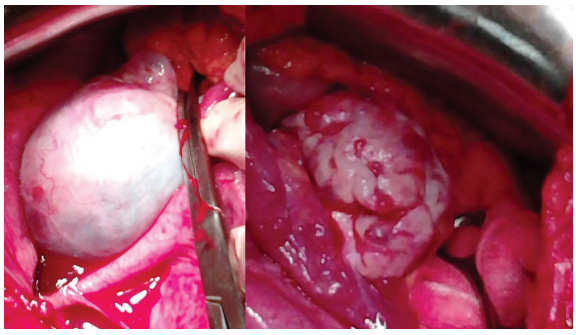

Fig. 3 Operative finding of left and right ovaries at 38 weeks shows slightly enlarged with multiple small left ovarian cyst and normal right ovary.

either as an accidental finding or in symptomatic pregnant women. Most of the cases are asymptomatic. The vast majority of HL arises in the third trimester but it can also occur in the first trimester[15]. A diagnosis of HL should be considered when multiloculated ovarian cysts are revealed during pregnancy especially in early gestation. Definite treatment is still controversial. Both conservative and surgical management were reported as successful management[4,6,14,16]. The decision for management should depend on the clinical presentation, the characteristics of the ultrasonographic finding, gestational age, and patient preference. Spontaneous regression has been reported[4]. Moreover, recurrence of HL in subsequent pregnancy has been reported[16].

Occasionally HL presents with edema, ascites, or pleural effusion that clinically mimics ovarian hyperstimulation syndrome[17]. The abnormal accumulation of fluid in patients with HL is caused by increased membrane permeability[15]. The presented case is the first case where HL was complicated by polyhydramnios without an identifiable cause. The authors hypothesized that the increase of membrane permeability may also occur at the placenta. The alteration of fetal membrane permeability associated with idiopathic polyhydramnios or oligohydramnios has been studied[18]. However, the exact mechanism that changes the membrane permeability cannot be clearly explained. A pregnancy complicated with HL should be monitored until delivery and puerperium. Polyhydramnios without an identifiable cause can put the patient at high risk for intrapartum and postpartum complications. However, common causes of polyhydramnios such as fetal anomaly, placental tumor, and maternal diabetes should be investigated.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Potential conflicts of interest

None.

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ครรภ์แฝดน้ำ ภาวะแทรกซ้อนพบได้น้อยในสตรีตั้งครรภ์ที่มีถุงน้ำรังไข่ชนิด hyperreactio luteinalis

ธารางรัตน์ หาญประเสริฐพงษ์, จิตติ หาญประเสริฐพงษ์

ถุงน้ำรังไข่ชนิด hyperreactio luteinalis ในสตรีตั้งครรภ์โดยธรรมชาติพบน้อย ครรภ์แฝดน้ำแทรกซ้อนในสตรีตั้งครรภ์ที่มีถุงน้ำรังไข่ชนิด hyperreactio luteinalis โดยไม่มีสาเหตุอธิบาย ไม่มีรายงานมาก่อน ในรายงานฉบับนี้ผู้นิพนธ์รายงานสตรีตั้งครรภ์ตัวอย่างซึ่งมีถุงน้ำรังไข่ชนิด hyperreactio luteinalis โดยพบขณะอายุครรภ์ 16 สัปดาห์ และได้รับการผ่าตัดเพื่อพิสูจน์ชิ้นเนื้อ หลังผ่าตัดไม่มีอาการแทรกซ้อน ขณะอายุครรภ์ 30 สัปดาห์ ตรวจพบว่ามีครรภ์แฝดน้ำ สตรีตั้งครรภ์คลอดทารกครบกำหนดแข็งแรงดี หลังคลอดถุงน้ำรังไข่ชนิด hyperreactio luteinalis ยุบลงได้เอง