Hyperreactio Luteinalis after Pregnancy with Severe Twin-to-Twin Transfusion Syndrome: A Case Report

Tikvica Luetic Ana, MD, PhD1*, Habek Dubravko, MD, PhD1, Nikolina Soken2, MD

1Department of Gynecology and Obstetrics, Clinical Hospital “Sveti Duh”, Catholic University of Croatia, Zagreb, Croatia
2Department of Gynecology and Obstetrics, University Clinical Hospital “Mostar”, Mostar, BiH

*Corresponding author: Ana Tikvica Luetic, Department of Gynecology and Obstetrics, Clinical Hospital “Sveti Duh”, Svetog Duha 64, 10000 Zagreb, Croatia, Tel: +385 98 573 159 (mobile); +385 91 37 12 261 (work), Email: ana_tikvica@yahoo.com, atluetic@kbsd.hr

Received: 01-15-2017
Accepted: 05-18-2017
Published: 05-25-2017
Copyright: © 2017 Ana Tikvica Luetic

Abstract

Hyperreactio luteinalis (HL) is a rare condition of bilateral cystic ovarian enlargement that usually develops in twin and molar pregnancies. Twin-to-twin transfusion syndrome (TTTS) is a severe complication of monochorionic twin pregnancies due to the pathological vascular anastomoses that often results with perinatal loss and serious morbidity. We present a case of patient with pregnancy loss due to the severe TTTS that later developed HL with complications of ascites and pleural effusion requiring active treatment. Conservative management with anticoagulant therapy was selected afterwards and the patient’s condition improved accompanied by decrease of ovarian size assessed by ultrasound. This case suggests that pathophysiology of HL could be related to TTTS.

Introduction

Hyperreactio luteinalis (HL) is a rare condition of bilateral cystic ovarian enlargement usually found in twin and molar pregnancies. The main cause of the condition is excessive ovarian stimulation by human Chorionic Gonadotropin (hCG) which is produced by the trophoblast with highest levels in the first trimester. Although HL has been clearly linked to elevated hCG levels the onset of the condition is variable pointing to their inconsistent relation [1]. Hyperreactio luteinalis is usually benign self-limited disease; however the appearance of the ovaries in the pregnancy suggesting malignancy has resulted in some cases with unnecessary operative procedures [2]. The condition is very rarely developed in otherwise low risk singleton pregnancies although there are published reports on HL in these pregnancies with complications such as fetal and maternal virilisation and severe preeclampsia [3, 4].

Twin-to-twin transfusion syndrome (TTTS) is a severe complication of monochorionic twin pregnancies that often results with perinatal loss and serious morbidity. The condition is result of the blood transfusion from the donor twin to the recipient twin through different vessel placental anastomoses [5]. It affects around 10% to 15% of twin pregnancies with one placenta while its early diagnosis followed intervention with placental laser ablation significantly improves the survival rate [6].

Here we present a case of severe hyperreactio luteinalis followed the pregnancy with TTTS treated by pleural puncture with later spontaneous resolution.
A 31-year-old secundigravida was referred to our department in the 19th week of pregnancy for additional surveillance due to the monochorionic diamniotic twin pregnancy. The patient had never taken drugs to induce ovulation. Ultrasound revealed twin-to-twin transfusion syndrome of third stage by Quintero. Donor twin was smaller with decreased body movements and oligohidramnios, while the recipient twin showed increased biometry measurements with polyhydramnios. The bladder could not be identified by ultrasound in the donor twin with additional periodically abnormal blood flow. Septostomy was performed with amnioreduction since the patient suffered from dyspnoea. Laboratory testing were normal with the exception of *E. coli* found in the urine sample followed by antibiotic treatment. Five days following hospitalization the patient complained of pains and bleeding and she spontaneously aborted dead fetuses of 410 and 210 grams with no visible malformations. Tissue samples of placenta, amnion and umbilical cord were positive for *Escherichia coli* and *Klebsiella pneumoniae* while cytogenetic testings showed both normal, male karyotype. Pathological examination revealed one placenta with artery-to-vein anastomoses and fetuses without malformations of internal organs. The patient was discharged the following day in good condition with normal vital functions.

The patient was readmitted to our department 10 days following abortion due to the extensive abdominal enlargement accompanied by dyspnoea and cough. Physical examination revealed distended abdomen up to chest line painful on palpation. Transvaginal ultrasonography showed a normal uterus with thin endometrial line but with bilateral enlargement of the ovaries (left ovary, 15x11 cm; right ovary, 16x10 cm) with multiple anechoic cysts that filled peritoneal cavity (Figure 1).

The ascites was abundant with fluid visible even in the upper part of the abdomen. Laboratory results included: leukocytes 13.23 x 10⁹, segmented neutrophil granulocytes 82%, lymphocytes 11.9%, C-reactive protein 96 mg/l, international normalized ratio 0.89%, D-dimers 8983 μg/l, albumens 35 g/l, human Chorionic Gonadotropin (hCG), 602.9 IumL, while the rest of the testing were normal. Chest radiograph revealed bilateral pleural effusion up to hilar line with interstitial oedema and elevated diaphragm. Pleural puncture guided by ultrasound was performed due to the significant symptoms of dyspnoea and cough and 700 ml of dark fluid was aspirated and analyzed without pathologic findings. Conservative management with anticoagulant therapy was selected afterwards. The patient’s condition improved daily with decrease of the abdominal circumference accompanied by balanced fluid intake and output. Repeated ultrasound surveillance showed reduction in the ovarian size measuring 7x6 cm on the right and 6x5 cm on the left side on the 10th day of hospitalization when patient was discharged home and followed later in outpatient clinic until the hCG negativization that occurred 2 weeks later.

**Discussion**

Hyperreactio luteinalis is a rare entity of ovarian enlargement due to the development of multiple simple cysts of unechogenic appearance with thin smooth walls expanded throughout the ovarian parenchyma (Figure. 1). This ultrasound feature is important not to be neglected due to the increased risk of complications such as preeclampsia, eclampsia, preterm delivery and other fetal and maternal possible risks [7]. This usually benign condition has to be distinguished from the ovarian malignancy which is also characterized by cystic formation but with thicker, irregular walls, solid areas and low impedance blood flow within the cyst. Precise ultrasound assessment could reduce a number of unnecessary surgical procedures done in a case when HL is misdiagnosed as potential ovarian malignancy. Another entity that has to be differentiated from HL is ovarian hyperstimulation syndrome which is complication of pregnancy conceived by assisted reproductive technology.
Hyperreactio luteinalis has been linked to the twin-to-twin transfusion syndrome by few authors [7, 8]. Takeda and co-workers have published a report of 4 cases of hyperreactio luteinalis associated with severe TTTS. They have detected 2 cases by ultrasound during pregnancy, one during caesarean section and one in postpartum. All the cases were diagnosed by ovarian enlargement, without serious maternal symptoms [8]. Our case was detected in the period after the abortion but with additional maternal complications such as abundant ascites and pleural effusion that resulted in dyspnoea and cough requiring active management. In retrospective study by Lynn and co-workers 31 patients with HL have been described. Although the authors analyzed only cases that were developed in prenatal period, TTTS has also been marked as associated factor of HL. Despite the fact that study included the largest number of cases with HL and complications such as: preeclampsia, pre-term birth and ovarian torsion have been described; no cases of maternal pleural effusion have been reported [7].

It is well known that the levels of hCG are elevated in twin pregnancy and many pathological condition of pregnancy that include placenta. Moreover, Hanaoka and co-workers have showed increased levels of hCG in a case of pregnancies complicated with TTTS especially of third and fourth degree by Quintero [9]. They also reported that effective laser surgery could result in decrease of serum hCG indicating that this hormone could be used for the assessment of laser ablation of vascular anastomoses in cases of TTTS. Described elevation of hCG levels in case of serious twin-to-twin transfusion syndrome could be influenced by the pathological changes of placenta and probably represents the causative link between LH and TTTS. Our case of serious TTTS was followed by additional serious HL, although in a time of diagnosis the hCG was not very high indicating the non-consistent relation between hCG levels and hyperreactio luteinalis incidence. The possibility of HL diagnose negligence during pregnancy in our case could not be rule out, but the period of 10 days post abortion without symptoms indicate the later occurrence of hyperreactio luteinalis. In addition, the massive enlargement of ovaries accompanied by maternal complications requiring pleural puncture is, to our knowledge, the first such case published in literature. Due to the low incidence of TTTS complicated by HL more cases with both entities will be hard to collect in order to elucidate the association between these two entities.

References


Cite This Article