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Incidental Finding Of Hyperreactio Luteinalis during Caesarean Section in Twin Pregnancy

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Abstract

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Keywords: Hyperreactio Luteinalis; Twin Pregnancy; Caesarean Section

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BACKGROUND: Some benign changes of the ovaries like hyper reaction luteinalis sometimes cannot be differentiated from malignant ones without histological examination. In these cases, surgical intervention sometimes cannot be avoided. Hyperreactio luteinalis is a condition that can occur only in pregnancy. It is characterised by bilateral benign multicystic ovarian enlargement.

CASE REPORT: We present a case of misleading intraoperative findings during Cesarean section that ended with ovariectomy.

CONCLUSION: During the Caesarean section, some benign masses of the ovaries, like hyper reaction luteinalis, are difficult to differentiate from malignant disease without histological examination, requiring surgical intervention.

Introduction

Hyperreactio luteinalis (HL) is a condition that can occur only in pregnancy. It is characterised by bilateral benign multicystic ovarian enlargement. The exact aetiology is unclear but high B-hCG or B-hCG – hypersensitivity is possible causes. It is very rare [1] [2]. Treatment is non-surgical, but sometimes it requires emergency surgery in case of ovarian torsion or haemorrhage [3].

We present a case of misleading intraoperative findings during Cesarean section that ended with ovariectomy.

Case Report

A 27-year-old patient in her first twin pregnancy was admitted to our department in the 31st

week of pregnancy for treatment of premature contractions. The pregnancy had been without complications to date and was followed up regularly. Standard laboratory tests were normal, and intravenous tocolysis was administered. Fetal ultrasound was normal, and a detailed ultrasound of the adnexa was not performed. The patient remained at our department up to her 33rd week of pregnancy, when her amniotic membrane ruptured spontaneously. The regular contraction started, and due to breech presentation of the first twin, delivery was performed by Caesarean section. The first twin was female, weighing 1780 g and 43 cm long, Apgar score 9/10. The second twin was male, weighing 1790 g and 44 cm long, Apgar score 9/10. During the Cesarean section surgery, multicystic and enlarged ovaries were identified, measuring 9 cm on the right and 8 cm on the left side (Figure 1 and 2). A frozen section biopsy of the right ovary was performed to rule out malignancy. This caused profuse haemorrhage that could not be controlled without ovariectomy. In the postoperative period, the patient had no complications. Histological examination of the right ovary revealed hyper reaction luteinalis. At ultrasound

examination, one-month post-surgery, the patient's left ovary was polyfollicular and measured 8 cm in diameter.



Figure 1: Intraoperative finding of right ovary

Discussion

Risk factors are multiple gestations, Rh sensitisation, twin to twin transfusion syndrome, gestational trophoblastic disease, hydropsfetalis, molar pregnancy, choriocarcinoma, polycystic ovaries, gestational diabetes, ovulation induction, clomiphene therapy, decreased the clearance of B-hCG due to renal dysfunction. The condition is most frequent in primiparas [4]. Hypothyroidism, PCOS, FSH secreting adenoma, or mutation in the FSH receptor can also lead to HL [5]. The genetic component in the development of the disease is excluded [1].



Figure 2: Intraoperative finding of left ovarii

Over 25% of cases of hyper reaction luteinalis are asymptomatic [6]. Symptoms are a low abdominal pain, nausea and vomiting, ascites, signs of virilisation, weight gain and shortness of breath[6].

Nausea and vomiting of pregnancy correlate positively with high levels of B-hCG [7]. Signs of virilisation are rare, occurring in only 30%of patients, mostly in their third trimester, and rarely in the second trimester [1], [2], [3], [4], [5], [6], [7], [8]. In our case, there were no clinical signs of maternal virilisation in pregnancy. Complications of this condition are preeclampsia, HELLP syndrome; fetal grow restriction, preterm delivery, torsion, dystocia and late onset of lactation in the post-delivery period. Remarkably high B-hCG levels might be predictive for preeclampsia as it may occur before 20 weeks' gestation [9]. Usually, it takes 2 months after childbirth for ovarian tissue to return to its normal size and ultrasonographic appearance [1]. Cases of virilized female fetuses are extremely rare [10], [11].

Diagnosis is mostly made by ultrasound, at a mean gestational age of 21.6 weeks with ovaries showing a characteristic "spoke-wheel" appearance. Finding of bilateral cysts, normal Doppler flow, and a lack of solid components differentiate HL from ovarian malignancies such as a borderline mucinous tumour of intestinal type [12], [13]. Laboratory findings may exhibit high levels of hCG, hyperandrogenism, and hyperthyroidism. The condition may be diagnosed incidentally during Cesarean sections and does not usually require any specific treatment. Adnexal masses can be identified in 0.3% of all cesarean deliveries, and most of them are incidental discoveries. In 96.7% of cases, the ovarian masses were characterized as benign, with only in 2.0% confirmed ovarian malignancies [14]. The rationale for surgical treatment is suspected malignancy, requiring histological evaluation through ovariectomy [15].

During the Caesarean section, some benign masses of the ovaries, like hyper reaction luteinalis, are difficult to differentiate from malignant disease without histological examination, requiring surgical intervention.

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