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BMH Med. J. 2021;8(4):149-152. **Case Report**

Spontaneous Hemoperitoneum in a Patient with Unscarred Pregnant Uterus: A Rare Case Report

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Introduction

Spontaneous hemoperitoneum in pregnancy (SHiP) - defined as blood within the peritoneal cavity of non-traumatic aetiology [1,2] is a rare, life-threatening event, particularly relevant to women with endometriosis if they are undergoing in vitro fertilization (IVF) treatment. This dramatic complication has been associated with high perinatal morbidity and mortality, but the trigger of the spontaneous bleeding has not been established [3,4]. Given the rarity of SHiP, its diagnosis is almost always unsuspected until the time of imaging, which is undertaken in patients who present with acute abdominal pain and/or distension and anaemia. Although the aetiology of SHiP remains unknown and may be multifactorial, several theories have been proposed to explain the rare complication.

Case Presentation

An elderly primigravida, at 27weeks of gestation (ICSI pregnancy with Donor egg) known case of hypothyroidism was referred to our tertiary care hospital complaining of a sudden onset of moderate lower abdominal pain since 2 hours with reduced perception of fetal movements in a state of shock. She denied trauma. The patient's past medical history was unremarkable. The course of the recent pregnancy had also been uneventful except with history of anaemia since second trimester on irregular iron supplements and on Ecospirin 75mg OD. Physical examination revealed tachycardia (148 bpm), BP of 80/40 mm Hg on adrenaline supports with severe pallor. An abdominal examination revealed that her abdomen was tense, distended and tender. The uterus was tense and tender, and fetal heart sound could not be localised with hand held Doppler. A gentle per vaginum examination after ruling out placenta previa was performed which revealed cervix soft, internal os closed, no bleeding p/v.

Counselling and Preparation

After obtaining high risk consent following an emergency USG scan suggestive of IUD and

moderate hemoperitoneum with Hb 6.4 gms, she was shifted to OR for exploratory laparotomy with wide bore canula inserted, necessary blood investigation done and multiple blood components issued with multidisciplinary approach alerting Senior Surgeons, Anaesthetists and blood bank. Patient's bystanders were counselled regarding the possibility of placental abruption or rupture uterus and need of laparotomy to rule out the same and the most likely complications like DIC, Sepsis, MICU admission, Massive blood transfusions, hysterectomy as well.

Revelation in OR

On opening the abdomen, she was found to have massive hemoperitoneum. Approximately 3 litres of dark colored altered blood and organised clots and the same was evacuated from the abdominal cavity, with distorted anatomy of uterus buried in adhesions. There were diffuse endometriotic implants all over the uterus which bled profusely. Bladder was pulled up and adherent to the anterior uterine wall, sharp dissection was done and adhesions were released, bladder was pushed down.A transverse incision was put in the lower segment, a fresh stillborn female baby as breech weighing 1080 gm was delivered. There was no evidence of abruption since the liquor was clear and no retroplacental clots seen. Uterus was closed in layers. Bleeding continued, Search for the source of bleeding continued and to our surprise, there were multiple rents, largest measuring 5 x 5 cm on the fundo-posterior wall and two lateral rents of 2 x 3 cm and 2 x 2 cm on the either sides with diffuse oozing. Repair of the rent was performed in two layers using vicyrl and hemostasis attained to some extent. Since the patient was on Ecospirin there was diffuse oozing. There were dense adhesions between sigmoid colon to the posterior surface of the uterus, and to the lateral pelvic wall, exteriorsation of the uterus could not be done. Senior Gastro-surgeon was called in for opinion and decided not to do the adhesiolysis since the patient was on Ecospirin and was in shock. With the support of uterotonics, transxemic acid and massive blood transfusion, abdomen was closed after keeping a drain in situ with catheter draining clear urine at the end of the surgery.

Critical Care and Supports

Post op period was uneventful managed in critical care unit with IV fluids, antibiotics, drain care (removed after 5 days) and serial monitoring was done to look for excessive bleeding She had total blood transfusion (7 units of PRBC, 4 units of FFP, 4 units of Platelet Concentrate). The patient was discharged from the hospital in good general condition after 11 days.

Counselling and education given to the patient and her family regarding avoiding future crisis pregnancy due to high probability of rupture uterus.

Discussion and Review of literature

Endometriosis and Deciduosis Are Associated with Decidual Bleeding

SHiP in patients with endometriosis raises the possibility of multiple bleeding sites in the pelvis. All of these need to be identified in order to achieve haemostasis. This can be very challenging, as the large uterus obstructs access to the posterior pelvic cavity during pregnancy. The posterior pouch of Douglas can also be obstructed by the presence of endometriosis-related adhesions and multiple endometriotic implants.

Aziz et al. [5] described a focus of endometriosis and decidualization in a woman who was 30 weeks into her first pregnancy and demonstrated the presence of very thin-walled blood vessels in the decidualized stroma. Mizumoto et al. [6] documented the presence of ruptured

vessels on the serosal surface of the uterine fundus at 20 weeks in a woman during her first pregnancy. Histology demonstrated an intense decidual reaction. The blood vessels were distended and exhibited disintegration of the vessel wall. Lier et al. described a case of late postpartum SHiP in a patient with ovarian endometriosis.

Mechanisms of Ectopic Decidual Bleeding

Emerging evidence suggests that endometriosis is associated with progesterone resistance characterized by the suboptimal expression of target genes [7]. Therefore, it is tempting to speculate that "functional" progesterone withdrawal triggers the involution of the decidual phenotype of the ectopic endometrium surrounding distended parametrial arterioles, leading to peritoneal bleeding of unpredictable severity.

In this case, there was evidence of endometriosis stage IV found during the laparotomy. A rupture of unscarred uterus is rare [8]. The ultrasound exam was beneficial in this case because it revealed the presence of free fluid inside the abdominal cavity and Intra uterine fetal demise, which led us initially to include uterine rupture in the differential diagnosis and minimised the delay in the decision to perform an emergency laparotomy to determine the site of bleeding, which could have been obstetric or non-obstetric. Rupture of the uterus should be considered in pregnant women with hemoperitoneum, even when caesarean section is absent from the obstetric history especially with menstrual diary suggestive of endometriosis.

Conclusion

Available evidence suggests that during pregnancy a link exists between ectopic decidualization, particularly that occurring in endometriotic foci, and the occurrence of SHiP. Alterations in vessels' walls have been demonstrated in the few cases where relevant biopsies were obtained and examined. Indeed, it seems that arterioles can become modified in the absence of trophoblast. There is a need to carefully examine vascular alterations at the site of bleeding leading to SHiP to gain information on the pathophysiology of this serious complication. Finally, there are indications that subclinical decidual bleeding may be a potential risk factor for preterm labour, further clinical, pathological and molecular investigations are required.

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