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Spontaneous Rupture of Sub-Serous Uterine Vessel in Advanced Gestation: A Rare Hemorrhagic Cause of Maternal Collapse- A Case Report

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1. Abstract

1.1. Background

Spontaneous rupture of an aberrant sub-serous uterine vessel is a rare but serious cause of maternal collapse during pregnancy, notably in advanced gestation. Spontaneous rupture of pelvic vessels is often misdiagnosed due to overlapping clinical presentations with other obstetric emergencies, such as placental abruption or uterine rupture. Literature indicates that spontaneous intra-peritoneal haemorrhage can arise from various sources, including ruptured utero-ovarian vessels and pelvic endometriotic deposits. The mechanism behind spontaneous vascular rupture remains poorly understood, though factors such as increased venous pressure and pre-existing vascular defects may contribute. Prompt diagnosis and intervention are crucial for maternal survival. This case report highlights the clinical presentation and management of a patient experiencing concealed intra-peritoneal haemorrhage due to this condition.

1.2. Case Report

We present the case of Mrs. W.L., a 41-year-old primigravida with a singleton gestation conceived via in-vitro fertilization (IVF). She presented with severe abdominal pain and hypotension, leading to a suspicion of concealed abruptio placenta at 36 weeks and 6 days of gestation. Initial examination revealed cold, clammy extremities and a blood pressure of 80/50 mmHg. Emergency resuscitation was initiated, and she underwent an emergency caesarean section and was delivered of a live female neonate with ligation of the bleeding vessel. Surgical findings included about 2 litres of hemoperitoneum and a ruptured aberrant sub-serous uterine vessel.

1.3. Conclusion

This case underscores the importance of maintaining a high index of suspicion for spontaneous rupture of pelvic vessels in pregnant women presenting with acute abdominal symptoms and signs of maternal collapse. Early recognition and surgical intervention are critical to prevent maternal morbidity and mortality. Enhanced awareness among healthcare providers regarding this rare complication can lead to improved outcomes for affected patients, particularly those with IVF pregnancies or prior histories suggestive of vascular abnormalities.

2. Introduction

Maternal collapse is a rare but potentially devastating event, and the outcome primarily for the mother depends on prompt and effective resuscitation, rapid diagnosis, and institution of a disease targeted therapy [1]. Maternal collapse is described as an acute event involving the cardiorespiratory systems and/or central nervous system, resulting in a reduced or absent consciousness level (and potentially cardiac arrest and death), at any stage of pregnancy and up to 6 weeks after birth. This may result from several causes, including absolute hypovolemia from hemorrhage, relative hypovolemia due to complication of spinal anesthesia, thromboembolism, amniotic fluid embolism, trauma resulting in cardiac tamponade, tension pneumothorax, and drug toxicity. Other causes include pre-eclampsia/eclampsia, and intracranial hemorrhage [1]. The main obstetric hemorrhagic causes include abruptio placenta, placental previa, ruptured ectopic gestation, uterine rupture, and rarely, spontaneous rupture of pelvic vessels has been reported. We report here-in a rare case of maternal collapse resulting from spontaneous rupture of an aberrant sub-serous uterine vessel.

3. Case Report

Mrs. W.L is a 41-year-old Gravida 1 Para 0+0 woman with in-vitro fertilization (IVF) conceived singleton gestation. She booked

index pregnancy in University College Hospital (UCH) at the gestational age of 26 weeks. Details of specific indication(s) for IVF were not known, except for primary infertility. Her baseline antenatal investigations were within a normal limit, and the syphilis, Hepatitis B, and Human Immunodeficiency Virus screening were also negative. She had no co-existing medical illness, and the pregnancy period remained uneventful until the gestational age of 36 weeks and 6 days, when she presented with 2 hours history of abdominal pain, which was initially localized to lower abdominal quadrants, but subsequently became generalized. She had a history of progressive body weakness, but bleeding per vagina or drainage of liquor. Her latest obstetrics scan revealed a live singleton intrauterine gestation in longitudinal lie and cephalic presentation. The estimated gestational age and fetal weight were 36 weeks and 2.7kg respectively. The amniotic fluid index was 10.2 cm, and the placental was in posterior-fundal location. She had no previous history of abdominal surgery, trauma to the abdomen. Findings on examination include a sweaty middle-aged woman, with cold, clammy extremities, and pallor. Her pulse rate was 128 beats per minute, and the volume was small. She was also found to be hypotensive with a blood pressure of 80/50mmHg. There was generalized abdominal tenderness, the fetal pole was difficult to palpate due to marked abdominal tenderness, and the fetal heart rate was 176 beats per minute. On vaginal examination, a normal vulva and vaginal was found, the cervix was posterior, medium in consistency, uneffaced, and the os was closed. The presenting part was at station 0-3. A suspicion of abruptio placental was made, and as such, immediate resuscitation with intravenous fluid, intranasal oxygen and a non-pneumatic anti-shock garment was applied. Consent for emergency caesarean section was obtained, and 4 units of blood and 2 units of fresh frozen plasma was prepared for her. She had emergency caesarean section and ligation of bleeding aberrant uterine vessel under general anesthesia. Surgical findings included; hemoperitoneum of about 2 L, and a uniformly enlarged uterus with well-formed lower segment and intact continuity of the uterine wall was noted. A live female neonate in cephalic presentation was delivered with a birth weight of 2.9 kg and an APGAR score of 6 at 1st and 8 at 5th minute of life. An actively bleeding ruptured tortuous vessel crossing over the uterine fundus from left cornual to the right was noted (Figure 1 and 2). Pouch of Douglas was obliterated by adhesions involving the posterior uterine wall, small and large bowel loops. She had two units of blood transfused. Her recovery from anesthesia and surgery was uneventful. The post transfusion packed cell volume was 28%. She was discharged on the fourth day after surgery.

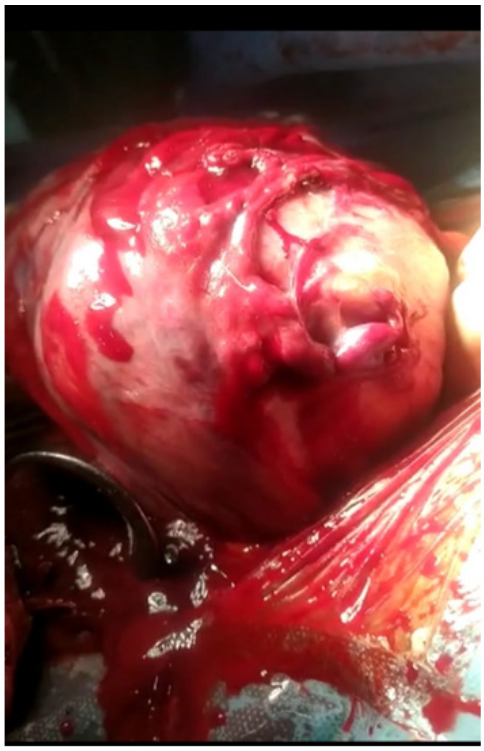

Figure 1: Uterine fundus with subserous vessels.

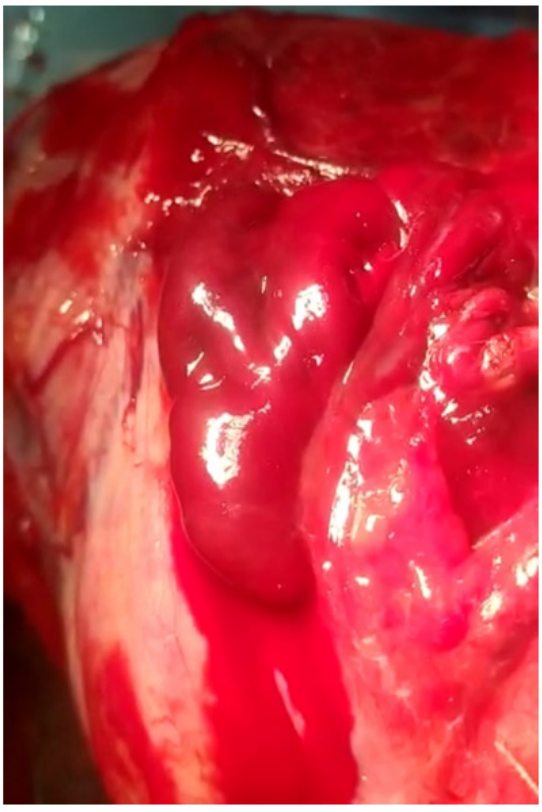

Figure 2: Bleeding ruptured subserous vessels.

4. Discussion

The case reported is a spontaneous rupture of an aberrant sub-serous uterine vessel in an advanced in-vitro fertilization (IVF) gestation, resulting in concealed intra-peritoneal hemorrhage. Intra-peritoneal hemorrhage due to rupture of aberrant uterine vessel may occur at any time in pregnancy [2]. Spontaneous rupture of pelvic vessel is a comparatively rare hemorrhagic cause of maternal collapse3-6. The common hemorrhagic causes of maternal collapse include; abruptio placenta, placental previa, postpartum hemorrhage, ruptured ectopic pregnancy, and uterine rupture. Some literatures have reported spontaneous rupture of pelvic vessels in advanced gestation as a cause of obstetric hemorrhage [3-6]. Just like in the case reported, the diagnosis is often missed due to several other surgical and obstetrical presentations that may pose a similar clinical picture. Though, an urgent bedside ultrasound scan could be a vital tool in the assessment and prompt management of the cases reported, this could not be done in the index case before surgery. The definitive diagnosis is established at emergency exploratory laparotomy, followed by caesarean section with ligation of the bleeding vessels to avert maternal mortality. The major causes of spontaneous intra-peritoneal hemorrhage from vascular rupture reported in literature include; ruptured utero-ovarian vessels, ruptured sub-serous uterine vessels, and from pelvic endometriotic deposits [7]. Some literature tried to establish a link between in-vitro conception, endometriosis, and the risk of rupture of uterine vessels resulting in spontaneous intraperitoneal haemorrhage [8]. Yan Zhang et al. [9] reported three cases of spontaneous intra-peritoneal hemorrhage among 573 women with IVF conception over a 3-year period. The common features in the 3 cases reported include; occurrence at 3rd trimester (29 to 35 weeks), prior diagnosis and/or treatment for endometriosis and pelvic inflammatory disease, the intra-peritoneal hemorrhage was secondary to spontaneous rupture of uterine varices/sub-serous vessels, and management required were; resuscitation, and proceed to emergency laparotomy for the cases. Lastly, preoperative diagnosis was missed and definitive diagnosis was established at emergency exploratory laparotomy. Similar cases of intraperitoneal hemorrhage were reported by Pillai et al. [9] and Vuong et al [10]. The precise mechanism of spontaneous vascular rupture is poorly understood. It has been proposed that the increased venous pressure in the uterine circulation and possible vascular defect acts synergistically to counter the effects of the physiologically hypertrophied vessel in pregnancy [11]. The vascular defect may result in quiescent aneurysmal or malformed blood vessels [11]. Conditions associated with hemodynamic stress, such as defecating, coughing, uterine muscular activity, may put pressure on already weakened vessels, leading to acute rupture and thus, hemorrhage. The 2 major gynecologic pathologies linked with these vascular defects are endometriosis [11,12] and pelvic inflammatory disease [11]. In the cases reported by Pillai et al, Vuong et al and Gomes et al., there was established pre-existing diagnosis of endometriosis [9,10,13]. The characteristic chronic inflammation associated with endometriosis and chronic pelvic inflammatory disease can lead to friable uterine vessels. Furthermore, the adhesions resulting from these pathologies, as seen in the index case reported, may cause further tension in these vessels as the uterus is enlarged during pregnancy [11]. In the case reported, the antenatal history of endometriosis and/or PID could not be established. However, findings of dense pelvic adhesion at laparotomy may imply the presence of either or both pathologies prior to conception. This may account for the antecedent history of infertility, and thus the need for in-vitro fertilization. It is recommended that cases with spontaneous intra-peritoneal hemorrhage in pregnancy should be managed by emergency laparotomy to ligate bleeding vessels irrespective of fetal condition to avert maternal mortality. Cesarean section should also be performed in instances where the uterine tension is high, there is any difficulty in achieving hemostasis, or if the fetus is non-viable, at term, late preterm or at other fetal conditions where chances of survival in the extra uterine life are favorable. If bleeding continues, or there is malformation of the uterus, hysterectomy should be performed. In the case reported, cesarean section was performed alongside ligation of the bleeding vessel, because the fetus had good chances of survival considering the gestational age at presentation.

5. Conclusion

Spontaneous rupture of pelvic vessel is rare, potentially life-threatening event, and an uncommon cause of maternal collapse. The presentation is non-specific, and as such, the pre-operative diagnosis is often missed and confused with abruptio placenta or uterine rupture. Women with IVF conception with antecedent history of endometriosis, and PID who presents with symptoms mimicking acute abdomen, and maternal collapse, there is the need for a high index of suspicion and good clinical judgement to avert the potential adverse obstetrics outcomes.

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