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Case Report

Obstetrics and Gynaecology Section

Ruptured Cervicoisthmic Pregnancy with 20 Weeks Intrauterine Foetal Death Lying in Left Broad Ligament- A Case Report

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ABSTRACT

Abnormal Low Lying Implantation of Ectopic Pregnancy (LLIEP) may occur in cervix, cervico isthmic region or caesarean scar. Cervicoisthmic pregnancy remains the rarest form of LLIEP, a life threatening cause of maternal morbidity and mortality with an incidence of 1 in every 2400 to 4500 pregnancies. In isthmic implantation, the gestational sac is located more cranially and between the cervix distally and the decidualised functional endometrium cranially. Transvaginal Ultrasound (TVS), Colour Doppler and Magnetic Resonance Imaging (MRI) remain gold standard modalities for early diagnosis. Ultrasonography depicts bulging lower uterine segment with a normal cervical length and consistency. Here, the author reports a rare case of ruptured isthmic pregnancy with 20 weeks intrauterine foetus death lying in left broad ligament in a 33-year-old unbooked $G_3P_2L_2$ with gestation of 31 weeks presenting in emergency. Ultrasonography depicted intrauterine foetus death with foetus lying in lower segment of uterus. Lower section caesarean section for failed induction confirmed hour glass uterus with empty upper uterine segment and bulging, distended, couvelaire lower uterine segment and left lateral rupture at cervicoisthmic junction. Dead 20 weeks foetus lying in leaves of left broad ligament. Peripartum hysterectomy was done as a life saving procedure. Cervicoisthmic pregnancy is rarest form of LLIEP and diagnosis may result in rupture with need of peripartum hysterectomy.

Keywords: Low lying implantation of ectopic pregnancy, Life threatening, Morbidity, Peripartum hysterectomy

CASE REPORT

A 33-year-old, unbooked $G_3P_2L_2$ at gestation of 31 weeks, muslim, homemaker, matriculate presented to Emergency Obstetrical Unit of tertiary care hospital with complaint of bleeding per vaginum and fever since last one day. There was history of bleeding per vaginum in late first and early second trimester. Patient did not perceive foetal movements throughout the pregnancy. Ultrasonography done two days back in private sector depicted intrauterine foetal death of 20 weeks gestation. Patient had no prior antenatal visits, nor had folic acid, iron or calcium supplements intake. Patient had two normal vaginal deliveries. Patient was average built with BMI 21 kg/m^2 with (Hb 8.4 gm%), with 110 bpm pulse rate, 100/70 mmHg blood pressure and 101.6 °F temperature.

On per abdomen examination, uterus was 20 weeks size, neither relaxed, nor tonically contracted however non tender with absent foetal heart sound. On per vaginum examination os was closed, posterior, uneffaced, long tubular with poor bishops score. A bedside ultrasonography reconfirmed intrauterine death at 20 weeks gestation, lower uterine segment was distended with foetus primarily in lower uterine segment, minimal free fluid in pelvis. Cervical ripening and induction was done with dinoprostone gel 0.5 mg eight hourly for three doses to which there was no response and even after oxytocin administration there was no onset of uterine contractions. On repeat pelvic examination, cervix was high up, 10-20% effaced, loose hanging, 3 cm dilated, presenting part felt with tip of finger which was soft placenta.

Patient was taken up for caesarean section under regional anaesthesia. There was a typical hour glass appearance of uterus, empty upper segment of uterus was sitting on top of over distended couvelaire lower uterine segment [Table/Fig-1]. There was left lateral rupture in cervicoisthmic junction. A dead foetus of 20 weeks gestation was lying transversely in leaves of left broad ligament while separated placenta was lying in lower uterine segment [Table/Fig-2]. Leaves of broad ligament were intact with minimal blood in peritoneal cavity. Tissues were foul smelling. Bladder serosa was congested with petechiae. A peripartum hysterectomy with repair of broad ligament haematoma was done. Cervix was normal in appearance with length of 3.5 cm [Table/Fig-3]. Massive blood and blood product transfusions was given to save the patient, 4 units Packed Red Blood Cells (PRBC), 4 units Fresh Frozen Plasma (FFP), 4 units platelet, 10 units cryoprecipitate was transfused. Patient was shifted to Intensive Care Unit (ICU) for 24 hours. Higher antibiotics, piperacillin and tazobactum 4.5 gm intravenous and metronidazole 500 mg intravenous eight hourly for 48 hours was given. Patient was discharged on postoperative day 11 under stable condition.

[Table/Fig-1]: Hour glass uterus with empty upper segment and bulging, distended, couvelaire lower uterine segment. **[Table/Fig-2]:** Twenty weeks dead foetus removed from left broad ligament and placenta removed from lower uterine segment. **[Table/Fig-3]:** Peripartum hysterectomy specimen with rupture at left cervicoisthmic region. (Images from left to right)

DISCUSSION

Abnormal LLIEP may occur in cervix, cervicoisthmic (close to internal os of the uterine cervix), and previous caesarean scars. Cervical pregnancy is a rare form of ectopic pregnancy and its incidence is about 1 in 1000 to 1 in 18000 live births. Caesarean scar pregnancy is another rare form of LLIEP with an incidence of 1 in 1800 pregnancies [1]. Cervicoisthmic pregnancy is located in an area limited by the histological internal os, at the junction of the endometrial and endocervical mucosae, and the anatomical internal os. Cervicoisthmic pregnancy is rarest of rare form of LLIEP with an incidence of 1 in every 2400-4500 pregnancies [2,3].

Cervicoisthmic pregnancy during the first trimester may present as abnormal painless vaginal bleeding, but carry the potential

risk of uncontrollable bleeding with less capability of muscular contraction and require time consuming resolution of the lesion with a prolonged recovery time. It may occur in women with a history of previous caesarean section. In first trimester, it is best diagnosed by transvaginal ultrasound and MRI [4].

Moreover, the combination of these two techniques allows better definition of disease evolution [5]. Ultrasonography typically shows an empty uterus and a gestational sac within the cervicoisthmic area, invading the anterior or the posterior wall of the cervix (hour glass uterus or dilated cervix). Colour Doppler imaging can be useful to confirm this diagnosis by identifying peri-trophoblastic blood flow. As soon as the diagnosis is confirmed, special care is needed. Based on the site, gestation age, size, and viability of the embryo/foetus, conservative treatment or pregnancy termination should be recommended [6].

Advancements in ultrasonography have led to the development of several conservative treatment approaches (medical or surgical) that avoid hysterectomy and preserve fertility. Medical methods have been developed more recently: intra-amniotic injection of potassium chloride or Methotrexate (MTX) and systemic chemotherapy with MTX. Surgical methods include uterine artery ligation and embolisation, Foleys catheter insertion, cervical curettage with or without circlage and more recently hysteroscopic resection [7,8]. Therefore, early diagnosis usually leads to early termination of pregnancy to avoid haemorrhage and hysterectomy. In some extremely rare cases like our case, this form of ectopic pregnancy may be discovered during labour and in majority of cases treated by a caesarean hysterectomy [9]. In the past few years, the diagnosis of aberrant gestation in the lower uterine segment is based solely on ultrasound findings rather than surgical and anatomic examinations [10].

During the first trimester, the cervix is limited cranially by the internal os which can be defined in two ways- histologically or anatomically. Histologically, the internal os is the transition point from the endocervical mucosa to the isthmic mucosa that resembles the corporal mucosa although thinner and richer of supporting tissue. The anatomical internal os which is the zone of transition between isthmus and uterine corpus is located 5-16 mm cranially to the histological os, therefore during the first trimester ultrasonography cannot distinguish the transition between isthmus and cervical canal but only between isthmus and uterine corpus. Two different hypothesis have been proposed for the origin of isthmic pregnancies. According to the first one, the gestational sac implants in the lower part of the uterine corpus with a subsequent extension of the implantation site into the isthmus and cervix [1]. An alternate hypothesis suggests that the original implantation occurs in the cervix and subsequently it extends above the internal cervical os into the lower uterine segment [3]. As per the latter hypothesis, the process resembles the normal implantation process which is with progressive incorporation of the lower segment of uterus into the gestational cavity [4], with one difference that it would begin from the cervix upward rather than from the cavity of uterus downward. Curettage, Asherman's syndrome, previous caesarean delivery, previous cervical or uterine surgery, and In Vitro Fertilisation (IVF) are aetiological factors for LLIEP [7]. Patients may present in first trimester with bleeding per vaginum [11]. In the present case also patient had episodes of bleeding per vaginum in late first and early second trimester. She did not have any antenatal visit or ultrasonography. Patient did not perceive foetal movements for which she was not evaluated.

Options for treatment of LLIEP depend on the general condition of the patient, gestational age at the time of diagnosis and the woman's desire to maintain fertility. Conservative management of cervicoisthmic pregnancy remains an option if diagnosis is made in early pregnancy. Local MTX chemotherapy or hysteroscopic resection have also been tried for conservative management [7]. While delay in diagnosis till third trimester may result in catastrophic complications with need of peripartum hysterectomy to save the woman. In the present case, the patient had to undergo peripartum hysterectomy in view of left lateral rupture at isthmus with placenta in lower uterine segment and dead 20 weeks foetus lying transversely in leaves of left broad ligament.

CONCLUSION(S)

Cervicoisthmic pregnancy is rarest of rare form of LLIEP, a life-threatening cause of maternal morbidity and mortality. It should always be kept in mind in case of first trimester haemorrhage. TVS, Colour Doppler and MRI remain gold standard modalities for early diagnosis and conservative management of cervicoisthmic pregnancy while delay in diagnosis till third trimester may result in catastrophic complications with need of peripartum hysterectomy.

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