



Case Report

A Cervico-Isthmic Pregnancy – A Categorically Catastrophic Ectopic Gestation

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Introduction

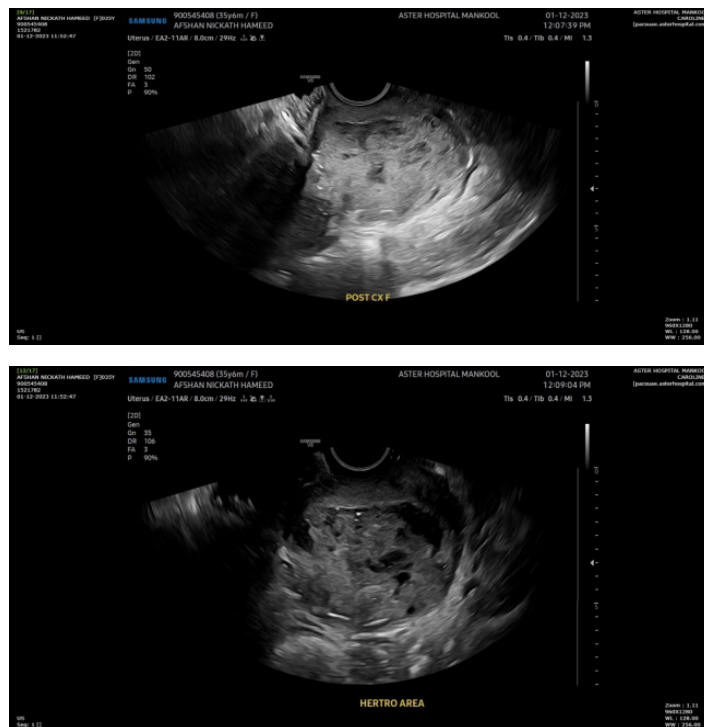
Cervico-isthmic pregnancy (CIP) is a rare and possibly life-threatening ectopic pregnancy in which the embryo gets implanted in the isthmus between the histological and anatomical internal os. The incidence of the CIP is 1 in 10 000 gestations. Over the last 40 years, only 19 reported cases that exceeded 20 weeks are available in the literature(11). If the pregnancies are detected earlier they are terminated due to the deadly complications that are involved. In certain cases, if the pregnancy continues beyond the first trimester it can lead to pernicious complications such as preterm birth, placenta previa, accreta spectrum, and life-threatening postpartum hemorrhage requiring peripartum hysterectomy. The pathogenesis involved which leads to the massive hemorrhage is due to the absence of contractility of the cervix and the isthmus. Here we present to you a case of G2a1 at 17 weeks of gestation presented to the emergency department with complaints of leaking per vaginum, post expulsion of the fetus we encountered massive life-endangering hemorrhage which was managed successfully.

Case report

25 yrs. G2A1 at 17+ 2 weeks of gestation, presented to the emergency department with complaints of pain and leaking per vaginum. On clinical examination, diagnosis of pre-viable Preterm Premature Rupture Of Membrane was confirmed. Ultrasound was done which showed funneling of the cervical canal for a width of 6.2mm and length of 7.4mm with effective cervical canal and absent liquor with the placenta in the fundus. She had undergone ultrasound (USG) at 12 and 16 weeks which showed a gestational sac in the endometrial cavity slightly on the lower aspect with normal cervical length of 3 cm but with funneling at 16 weeks. She was admitted for termination of pregnancy after explaining to the couple the procedure and the risk of bleeding. Under

antibiotic cover priming of the cervix was done and enhancement of miscarriage was done with misoprostol and carboprost. She developed a moderate amount of bleeding per vaginum, hence after discussing with the patient and her partner, she was taken for evacuation of the products of conception under general anesthesia. During the procedure the fetus was in the vagina, same was removed using sponge holding forceps, post removal, she had profuse bleeding per vaginum with placenta in situ, uterotonics were administered and proceeded with removal of retained products. 1.5 liters of initial blood loss was estimated and a massive hemorrhage protocol was initiated .Multi-disciplinary team was involved senior anesthetist, vascular surgeon, intensivist and radiologist. Intraoperative ultrasound was done to ensure the completeness of evacuation and locate the source of hemorrhage, which showed that the bleeding was not from the fundus, it was from the lower uterine segment and cervix, which confirmed the implantation of pregnancy at the cervico – isthmic region (retrospective diagnosis of cervico- isthmic pregnancy) FOLEY’S bulb was inserted and tried balloon tamponade but in vain as bleeding continued. Hence proceeded with laparotomy due to persistent heavy bleeding with total blood loss estimated at around 2.5 litres, simultaneously patients partner was alerted regarding the maternal condition and the possibility of hysterectomy. The abdomen opened in layers, multiple compression sutures were taken on the uterus and CERVIX due to persistent bleeding. We proceeded with step wise devascularization, uterine arteries ligation followed by internal iliac artery ligation. Total blood loss of 4 liters was estimated. Hemostasis was achieved following internal iliac ligation. Multiple blood and blood products were transfused. After observing for 30 minutes for vaginal bleeding, abdomen was closed with intra - abdominal drain, shifted to the ICU for further care. Thus after a Herculean effort we succeeded in conserving her uterus and preserved her fertility.

Postoperative period was uneventful, she was discharged from the hospital on post-operative day 5 and she came for follow up after a week. Post op usg was done after 3 weeks which revealed heterogenous echoes in cervix, with negative beta-hcg which again confirmed the diagnosis of cervico isthmic pregnancy. She was advised to avoid conception for the next 6 months and insisted on pre-conceptional assessment.



Post-operative Images- Confirms a Cervico Isthmic Pregnancy

Discussion

In 2013, the Tsai group introduced a new terminology of low-lying implantation ectopic pregnancy (LLIEP), which refers to cases where implantation occurs near the cervix and has been clearly classified into Cervical Pregnancy, Caesarean Scar Pregnancy, and Cervico- Isthmic Pregnancy Cervico-isthmic pregnancy is a rare complication of sac implantation, which is poorly characterized both diagnostically and in its development. The internal os is defined histologically as the transition area from the endo-cervical mucosa to the isthmic mucosa and anatomically as the transition area between the isthmus and uterine corpus [1-3] which is located 5 to 16 mm cranially to the histological os. Therefore, during the first trimester, ultrasonography cannot distinguish the transition between the isthmus and cervical canal, but only between the isthmus and uterine corpus. From the 12th week onward, the isthmus progressively unfolds into the uterine

cavity, becomes occupied by the gestational sac, and takes the name of the lower uterine segment. Normally, the gestational sac implants on the uterine corporal decidua, and only during the second trimester does it occupy the space of the widened isthmic canal. Gestational products in CIP extend to the lower uterine segment; therefore, they can grow until the fetus is viable. If it progresses to the second trimester it can lead to pernicious complications such as placenta previa, placenta accreta spectrum, postpartum hemorrhage requiring hysterectomy, intensive care unit (ICU) care even death if not treated effectively. Keeping the complications in mind which are catastrophic, if CIP is diagnosed in the first trimester termination of the pregnancy by methods including methotrexate (MTX) injection followed by suction evacuation can be considered. As CIP is a form of ectopic pregnancy, termination is still the treatment of choice. Any pregnancy in the lower half of the uterine cavity has to be considered as cervical isthmic pregnancy and meticulous evaluation, follow up and prompt treatment in a well-equipped hospital is mandatory.

True cervical pregnancy is defined by its location completely within the cervical canal, with no placental tissue above the internal os (3, 4), and an intact part of the cervical canal between the gestational sac, distally, and the uterine decidua, proximally (1). In cervico-isthmic implantation, the gestational sac is located more cranially, and the gestational sac is located between the cervix, distally, and the decidualized functional endometrium, cranially. Clinical examination can confirm the ultrasonographic diagnosis by detecting a barrel-shaped hyperemic cervix in the presence of a cervical pregnancy, and a normal cervical length and consistency with a bulging lower uterine segment in the case of an isthmic pregnancy.

Two different hypotheses have been proposed for the origin of cervical-isthmic pregnancies, the first one is that the gestational sac gets implanted in the lower part of the uterine segment, with a subsequent extension of the implantation site into the isthmus and cervix (5, 6). Another alternative hypothesis suggests that the original implantation occurs in the cervix, and thereafter it extends above the internal cervical os into the lower uterine segment (4). According to the latter hypothesis, the process could resemble the normal implantation process, which is the progressive incorporation of the lower uterine segment into the gestational cavity (2), with the difference that it would start from the cervix upward rather than from the uterine cavity downward. Independently from the causative hypothesis, a weakness of the internal cervical os has been claimed to play a role in the pathogenesis of cervico-isthmic pregnancies.

The criteria for a prenatal sonographic diagnosis of cervico-isthmic pregnancy are not clear, owing to the rarity of the condition. Often the condition is described clinically and sonographically as a cervical pregnancy, but it is later on defined as cervico-isthmic

in the face of a successful conservative management until fetal viability, which is thought impossible for true cervical pregnancies (4, 7).

If the pregnancy is identified in the early first trimester termination of pregnancy should be considered, especially if there is a desire for fertility preservation. In such cases the management is similar to cervical pregnancies with methotrexate (systemic or intra-amniotic), surgical evacuation with balloon tamponade, hysteroscopic resection and uterine artery ligation or embolization. If the pregnancy crosses second trimester and further on, adequate counseling and strict follow up of the patient, and expectant management at the time of labour is needed

In our case cervico isthmic location of pregnancy was suspected and confirmed intraoperatively due to the massive hemorrhage encountered. Hence it was a retrospective diagnosis.

Conclusion

The rarity of CIP makes it a diagnostic challenge. It is a potentially life and fertility-threatening condition, hence early and prompt diagnosis followed by care of pregnancy in a tertiary center. With involvement of experienced obstetrician, multidisciplinary team can make difference in the life and fertility of a woman.

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