

Case Report**Miracle baby: Very rare case of heterotopic pregnancy with cervical ectopic in elderly primigravida with very poor ovarian reserve with oligoastheno-terato-zoospermia conceived with IVF**Saraogi RM¹, Yadav P², Poddar P³, Chavan K⁴

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ABSTRACT

A 42 year old with primary infertility over 2 years with poor ovarian reserve (AMH 0.02) with an Oligoastheno-terato-zoospermia had 3 attempts of embryo transfer with her own egg. USG showed a heterotopic pregnancy with fundal 5-6 weeks intrauterine gestational sac with a cervical ectopic with small subchorionic collection along with posterior uterine wall. Despite embryo reduction, it still showed a persistent heterotopic pregnancy with an increasing subchorionic collection in the cervical canal. There was PROM at 27 weeks and under adequate antibiotic and steroid cover, classical caesarean section was done. Due to an adherent cervical pregnancy remnant, bilateral internal iliac artery ligation with an obstetric hysterectomy was done. As few cases exist in literature, there are no specific and no universally accepted treatment modality for heterotopic cervical pregnancy and treatment depends on experience, equipment of medical team and maternal conditions.

Keywords: Ovarian reserve, heterotopic pregnancy, embryo-reduction, obstetric hysterectomy

Introduction

Heterotopic pregnancy, the presence of two gestational sacs simultaneously, is a rare event but with the advent of Assisted Reproductive Technology, it is now an increasingly common complication. The most common combination is an intrauterine and an extra uterine gestation, most of which are in the tube (90%), but implantation in the cervix, ovary, interstitial segment, abdomen, and previous caesarean scar have been reported. ^[1] The low incidence of the entity and improving

treatment options necessitate this rare condition to be discussed repeatedly.

Case History

A 42 year old female married over 2 years approached our hospital as a case of primary infertility with a poor ovarian reserve (AMH 0.02) oligo-astheno-terato-zoospermia in husband and underwent ART Treatment and successfully conceived after three cycles of embryo transfer with her own egg on 19.9.2014. Her pregnancy test was positive and she was started on

progesterone, folic acid, aspirin and hypothyroid medications. Serial Ultrasonographies for fetal viability were done.

At 5 weeks, USG was suggestive of an embryonic pole with fetal cardiac activity + ~ 5-6 weeks CRL 3 mm with small intrauterine SOL. A repeat USG showed a heterotopic pregnancy with a cervical ectopic and fundal intrauterine live gestation with subchorionic bleed. Aspirin was stopped and while continuing progesterone supplementation, USG guided embryo-reduction was done at 11 weeks. However despite this, it still showed a persistent dichorionic twin pregnancy with single intrauterine fetus with persistent subchorionic small hematoma and regressed embryo with enhanced peripheral vascularity in the posteriorly placed placenta. She was kept on close observation and adequate progesterone was given.

At 20 weeks, patient presented with a single episode of spotting PV and was admitted for observation and evaluation and treatment. A repeat USG showed reduced twin in the endo-cervical region (sac volume 48 cc) with presence of enhanced blood flow with a live intra-uterine gestation sac of 21.1 weeks. APLA, ANA levels were within normal limits.

At 23 weeks, patient complained of passage of 80-100 cc clots per vaginum. An urgent USG revealed a sub-chorionic collection increased to 403 cc along the posterior uterine wall. Reduced twin tissue volume was also increased to 161 cc with blood flow within.

At 25 weeks, patient developed high grade fever with chills with UTI and leucocytosis and a raised procalcitonin and CRP levels. She was started on broad-

spectrum antibiotics including carbapenems and infection was controlled. During this course, she was continued with tocolytics (isoxsuprine), progesterone analogues, complete bed rest, intravenous iron sucrose supplementation. Two doses of intramuscular steroid were administered 24 hours apart. She was monitored daily and a careful watch on bleeding was done. However after 2 weeks, she complained of leaking PV since 3 AM with prominent uterine contractions and. an urgent USG showed a 27.3 weeks intra-uterine gestational sac with severe oligohydramnios and transverse lie and EFW being 1150 gm and cervical tissue with a rich vascular supply and a sub-chorionic hematoma of 40 cc and funnelling of lower uterine segment with cervical length being 1.6 mm. After a high risk consent and keeping adequate blood and blood products ready, she was taken up for an emergency caesarean section. To our shock and dismay, there were fetal remnants of cervical pregnancy found adherent to cervix and lower uterine segment. All attempts to remove the remnants were in vain and caused profuse haemorrhage. A classical caesarean section was done and delivered a 920 gm male baby who cried immediately after birth and was admitted in NICU. Placenta was removed spontaneously. Since there was presence of profuse haemorrhage from the adherent cervical fetal remnants, a mandatory decision to do an obstetric hysterectomy was done followed by right internal iliac artery ligation. Postoperative patient was stable and received 2 units packed cells. Mother was sent home when stable and baby was in NICU for further preterm management. Both the baby and mother have been doing well and are under regular follow-up.

Discussion

Heterotopic pregnancies are relatively very rare complications of pregnancy, incidence being estimated at 1 in 30,000 pregnancies. [2] Over time, there has been a considerable increase in its incidence owing to pregnancies following various infertility treatments especially ovulation induction. An even greater incidence of around 1 to 3 per 100 pregnancies is reported with assisted reproduction techniques (ART) like In-vitro fertilization (IVF) and gamete intra-fallopian tube implantation (GIFT). A further increase is reported with 4-embryo transfers of 1 in 45 pregnancies.

A cervical heterotopic pregnancy is altogether a very rare event with most such cases being reported after infertility treatment. Cervical pregnancies usually abort early in the first trimester however if they are implanted in the cervico-isthmic region or more towards the uterine cavity, the chances of continuation of pregnancy is even higher. Cervical pregnancies were previously complicated due to late diagnosis due to non-availability of ultrasonogram and were usually detected during dilatation and evacuation complicated by torrential bleeding requiring hysterectomy in most cases. However with the advent of high resolution ultrasonogram and Magnetic resonance imaging (MRI), it is usually detected at an earlier stage and hence better managed. Placental implants in the cervical region, internal echoes within an enlarged cervical canal, and hour-glass shaped uterus are suspicious of cervical pregnancy. A color Doppler with a high peri-cervical and peritrophoblastic blood flow helps in differentiating a cervical pregnancy from an aborting uterine pregnancy.

In general, the aims of the treatment are the protection of a coexisting intra-uterine pregnancy (IUP), the minimization of blood loss, and fertility preservation. Since heterotopic cervical pregnancy is a rare entity, there are not established standard protocols but several options exist for the management. Operative management is still a mainstay, but it involves surgical and anesthetic risk to both the mother and fetus. Uterine artery ligation or angiographic arterial embolization may result in the radiation of the viable IUP, and influence on endometrial receptivity, which could decrease future fertility. [3] Embryo reduction procedure by ultrasound-guided transvaginal injection of potassium chloride into the ectopic component is another treatment modality. [4] The ongoing pregnancy might be complicated by persistence and even enlargement of remaining trophoblastic tissue, leading to obstetric hemorrhage and emergency intervention. Local methotrexate administration may be used with the known risk of systemic adverse effects such as thrombocytopenia, leukopenia, elevated serum liver enzymes, and especially the teratogenic effect, taken into consideration. [5] Systemic methotrexate administration may be considered only when the aim is not to preserve the IUP.

Surgical treatment may include suction evacuation, cervical curettage with or without cerclage and Foley catheter insertion. Foley catheter insertion and cervical cerclage seem universal salvage maneuvers to stop early or late bleeding whichever technique is used to terminate the cervically located gestation. [6] Nonsurgical treatments may not be rationale in women who are hemodynamically unstable. Laparotomy

may rarely be needed if any even in women with profound bleeding. For cervical pregnancies without a simultaneous intrauterine gestation, complete evacuation of the pregnancy or systemic methotrexate administration seem better options due to the risk/benefit ratio of the reduction procedure.

In our case, since the patient was hemodynamically stable and the aim was to conserve the IUP, embryo reduction could be done after counseling the risk of the potential postoperative complications including bleeding, abortion of the IUP, cervical mass infection which may lead to premature rupture of the membrane, and postpartum bleeding and severe bleeding leading to potential need for emergency procedures including even hysterectomy.

Conclusion

With no definite treatment modalities, it poses as a challenge to the obstetrician and management needs to be individualized according to the hemodynamic status, technical availability of the facility and skills of the surgeon. But more important, is keeping suspicion of heterotopic pregnancy in mind and an early diagnosis with the recent advanced treatment modalities.

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