

# International Journal of Clinical Obstetrics and Gynaecology

ISSN (P): 2522-6614  
ISSN (E): 2522-6622  
© Gynaecology Journal  
[www.gynaecologyjournal.com](http://www.gynaecologyjournal.com)  
2021; 5(4): 228-231  
Received: 19-05-2021  
Accepted: 21-06-2021

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## Revamping rarity: Successful treatment of heterotopic cervical pregnancy with transvaginal guided cervical aspiration and intra-amniotic KCL instillation: Case reports

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DOI: <https://doi.org/10.33545/gynae.2021.v5.i4d.991>

### Abstract

**Objective:** To present case reports of heterotopic cervical pregnancy successfully treated with conservative transvaginal ultrasound guided aspiration followed by intra-amniotic instillation of KCL.

**Design:** Case report

**Setting:** Advanced ART – ICSI cycle

**Patients:** Two cases of infertility with cervical heterotopic pregnancy following ICSI FET cycle.

**Intervention:** Transvaginal ultrasound guided intra-amniotic aspiration followed by intra-amniotic instillation of KCL.

**Main outcome measure:** Successful outcome of intrauterine pregnancy, decreased maternal and fetal morbidity and recovery of the patients.

**Result:** Successful selective reduction of cervical pregnancy and delivery of the intrauterine pregnancy at term.

**Conclusion:** Transvaginal ultrasound guided aspiration followed by intra-amniotic instillation of KCL can be safely used as a conservative measure to manage cervical heterotopic pregnancy.

**Keywords:** heterotopic pregnancy, cervical pregnancy, transvaginal ultrasound, selective reduction, intra-amniotic KCL injection

### Introduction

Rarity includes curiosity. Ectopic pregnancy is always a nightmare for any obstetrician. When it becomes precious as seen in heterotopic pregnancy where an intrauterine pregnancy coexist with pregnancy elsewhere, in an ICSI cycle, then the definite need for salvage arises. Ginsburg ES et al in 1994 [1] reported first live birth of cervical heterotopic pregnancy following transvaginal reduction with KCL. Since then cases of cervical heterotopic pregnancy and their conservative management though reported in literature, is still a rare entity. In 2007 Shmuel nitke *et al.* [2] reported a case managed with intraarterial instillation of methotrexate followed by embolization of uterine arteries but intrauterine pregnancy cannot be salvaged. In 2002 Mashiach *et al.* [3] performed shirodkar cerclage for 8 wks heterotopic pregnancy with evacuation of cervical pregnancy followed by term delivery of intrauterine fetus. In 2003 Jozwiak *et al.* [4] did hysteroscopic resection of cervical heterotopic pregnancy followed by term delivery. Several case reports for using intrafetal Kcl [5, 16] has been published. As methotrexate has its own adverse effects and surgical procedures may lead to catastrophic hemorrhage and might warrant even hysterectomy we opted transcervical intra fetal instillation of KCL followed by aspiration. Here we report three cervical heterotopic pregnancies gone in three different ways and their successful outcome. All three patients were properly counselled regarding the pros and cons of the procedure and considering the preciousness patients opted for reducing the cervical heterotopic pregnancy. Proper consent got and proceeded.

### Case reports

#### Case Report 1

A 32 year old woman, married 3 yrs came to us as a case of primary infertility. The couple underwent four IUI cycles outside and a diagnostic hysterolaparoscopy as her screening HSG

showed bilateral tubal block. Her AMH was 3.4 and husband's semen parameters were normal. Her follow-up scans showed an endometrial polyp. Diagnostic hysteroscopy was done. Polypectomy with endometrial curettage was done for polypoidal endometrium. As her internal os was very tight os dilatation was done with no.7 hegar's. Empirical AKT given for 6 months as endometrium TB PCR was reactive. ICSI cycle with fresh transfer has been planned for her.

GnRH agonist long protocol was followed. 9 days stimulation with FSH and HMG done and hCG trigger was given on day 10 and ovum pickup done 35 hrs later. 20 oocytes were got in total with 16 metaphase II oocytes and 8 were fertilized (2 morula, 1 6CG I, 3 4CG II, 2 3CG I). She had a fresh transfer of 3 day 3 embryos (2 morula, 1 6CG I). Luteal phase support with 50mg / day intramuscular progesterone and 400 mg/day of vaginal micronized progesterone and 6 mg/ day of oral estradiol valerate was given. After 12 days of embryo transfer serum  $\beta$ hCG level measured which showed a positive value of 932 m IU/ml and a 48 hour doubling value was 1680 m IU/ml. she had her first scan after a week which showed a single intrauterine sac with yolk sac and a small sac like structure near cervix. Her follow-up scan after a week at 6 weeks showed a cervical heterotopic pregnancy with a intrauterine sac with yolk sac and fetal pole with good cardiac activity and another sac with yolk sac and fetal pole with cardiac activity in cervix.



**Fig 1:** Gestational Sac with Yolk Sac in Cervix



**Fig 2:** Combined Intrauterine and Cervical Sac with Yolk Sac and Fetal Pole

Since our case is an infertile couple with unexplained infertility and finally achieved success with ICSI and as it is a precious pregnancy, it has been a great challenge for us to save the

intrauterine pregnancy. On discussion with a fetal medicine specialist and expert team selective fetal reduction of the cervical pregnancy planned. We opted medical management by transvaginal ultrasound guided aspiration of intra-amniotic fluid followed by intra-amniotic KCL injection.

Patient was prepared for the procedure. Emergency surgical team was available standby to intervene any catastrophe if happens. IV sedation was given and a 17 gauge aspiration needle was used and through transvaginal approach the needle was inserted using a biopsy guide and the cervical sac was reached and the intra-amniotic fluid was aspirated and 3 ml KCL was injected intra amniotically. After 5 minutes cardiac was checked which was absent. She was discharged after one day of observation. On discharge intrauterine sac's cardiac activity was reconfirmed.



**Fig 3:** Transvaginal Reduction of Cervical Pregnancy

The patient has been followed frequently and was given high risk obstetric care throughout pregnancy with serial infection screening. The pregnancy continued till 37 weeks uneventfully. Emergency caesarean section was done as patient had preeclampsia. The patient delivered a healthy female baby of birth weight 2.8kg but had torrential bleed from the cervical site following delivery and underwent caesarean hysterectomy with massive blood transfusion of 5 units PRBC and 3 units FFP both intraoperatively and postoperatively. The patient was monitored in ICU for 5 days and had been discharged on 10<sup>th</sup> postoperative day.

### Case Report 2

Mrs X, 36 yrs, married 6 months (second marriage), with history of one spontaneous abortion in first marriage came for infertility treatment. Infertility workup done and her AMH was 1.9 and semen parameters were normal. As her outside hysterosalpingogram showed bilateral tubal block diagnostic hysterosalpingography done. Bilateral salpingectomy done as she had bilateral hydrosalpinx followed by which ICSI FET cycle done and two day 5 blastocyst transferred. Beta hcg positivity and doubling noted after 12 days. Clinical pregnancy confirmed at 5 weeks by transvaginal ultrasound where cervical heterotopic pregnancy was diagnosed with one intrauterine sac with yolk sac and one sac in cervical canal. Follow up scan a week later showed viable sac both intrauterine and cervical. We opted medical management by transvaginal ultrasound guided aspiration of intraamniotic fluid followed by intraamniotic KCL



injection. The procedure carried out as in the first case. Unlike the first case in this case post reduction there was remnant cervical mass which was gradually increasing in size. The pregnancy was carefully monitored and we did elective cesarean section at 38 weeks as the patient had pain and delivered a healthy male baby of birth weight 3.0 kg with a surgical team standby. Luckily there was no undue bleeding and a thick white slough like mass about 3x2 cm seen in the posterior lower segment extending to the cervical canal noted and as it was densely adherent no attempt was made to remove it. Postoperatively follow-up scan on 4<sup>th</sup> postoperative day showed the cervical mass reduced in size with no vascularity. The patient got discharged the next day and follow-up 15 days later showed a near normal cervical canal.



**Fig 4:** Intrauterine Pregnancy with Cervical Mass



**Fig 5:** Homogenous Mass in Cervix



**Fig 6:** Increasing Size of Cervical Mass



**Fig 7:** Remnant Slough Near Cervix

### Discussion

Cervical pregnancy is rare and if left untreated is a life threatening condition. The incidence is 1 in 1000 to 1 in 18000 pregnancies. Heterotopic pregnancy is when an intrauterine and extra uterine pregnancy coexist. The incidence being 1 in 30000 pregnancies commonly combined intrauterine and tubal pregnancy. Cervical heterotopic pregnancy is an extremely rare condition. The incidence being increased with assisted reproductive techniques.

The etiology being unclear, may include cervical abnormalities, prior surgical procedures like dilatation and curettage, caesarean section or reflux of embryo during embryo transfer, cervical trauma during embryo transfer. Due to increased vascularity and no muscular elasticity in cervix, cervical pregnancy can lead to catastrophic haemorrhage and even hysterectomy.

The advancements in transvaginal ultrasound [7] had resulted in early diagnosis and give a chance of successful conservative management being diagnosed early. Ultrasound diagnostic criteria of cervical pregnancy include

- Gestational sac or placental tissue visualized within the cervix
- Cardiac motion noted below the level of internal os
- Hourglass uterine shape with ballooned cervical canal
- No SLIDING SIGN, no movement of sac with pressure from transvaginal probe

The challenge in heterotopic cervical pregnancy is the desire to maintain the intrauterine pregnancy. Review of literature guided us with various conservative treatment modalities on selective fetal reduction of cervical pregnancy using both surgical and medical management. In 2019 Sindhubairavi *et al.* [8] reported a case managed by transvaginal ultrasound guided aspiration of the products followed by successful outcome as in our case. In 2004 Gyamfi C *et al.* [5] reported a cervical heterotopic pregnancy reduced with transvaginal instillation of KCl followed by aspiration and complicated by catastrophic antepartum hemorrhage at 31 weeks undergoing hysterectomy. In 2012 Ji Won Kim *et al.* [9] reviewed various treatment modalities. Surgical excision of cervical pregnancy were done by various techniques like aspiration, extraction with forceps, cervical curettage followed by foley catheter insertion to tamponade the haemorrhage, electrocauterisation [10] of bleeding points, cervical stay suture [11] of lateral cervical vessels and cervical cerclage following surgical excision. Medical management includes intraamniotic instillation of methotrexate [12], hypertonic

glucose, hypertonic saline, KCL. Uterine artery embolisation<sup>[13]</sup> before surgical excision can prevent bleeding during and after procedure. Considering the preciousness and priority to save the intrauterine pregnancy we opted conservative medical management with combined aspiration with intraamniotic KCL injection.

### Conclusion

Thus heterotopic cervical pregnancy though an extremely rare and life threatening, the recent advances in early diagnosis and conservative medical management with expert team and monitoring in high risk suite can give a fruitful result without compromising maternal and fetal safety. In our cases conservative intervention done and one ended in term delivery followed by hysterectomy and the other with term delivery with no much complications.

### Compliance with Ethical Requirements

#### Conflict of Interest

The authors declare that they have no conflict of Interest.

#### Ethical Statement

Hereby, I Dr. Pradeepa Sudhakar consciously assure that the for the manuscript submitted above where in accordance with the ethical standards and informed consent was obtained from all the patients being included in the study, Prior ethical approval was obtained from the Sudha Hospital Ethical Committee, Coimbatore, Tamil Nadu.

#### Informed Consent

Informed consent was obtained from all the patients for being included in the study

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