Tropical Journal of Obstetrics & Gynaecology
ISSN-Print: 0189-5117
Online: 2543-148X
Official Publication of Society of Obstetrics & Gynaecology of Nigeria

Case Report

Successful Management of an Intrauterine and Cervical Heterotopic Pregnancy: A Case Report

***Aniekan M. Abasiattai, *Ntiense M. Utuk, *Iniobong Ettete, **Felix Uduma, ***Otu Etta, *Aniefiok J. Umoiyoho, *Ubong Mbaba**

**Department of Obstetrics/Gynaecology, University of Uyo Teaching Hospital, Uyo, Nigeria*
***Department of Anaesthesia, University of Uyo, University of Uyo Teaching Hospital, Uyo, Nigeria*
****Department of Radiology, University of Uyo Teaching Hospital Uyo, Nigeria*

Corresponding Author

Professor Aniekan M Abasiattai
Dept. of Obstetrics/Gynaecology
University of Uyo Teaching Hospital,
PMB 1136, Uyo, Nigeria
Email:animan74@yahoo.com

ABSTRACT

Heterotopic cervical pregnancy is an extremely rare and potentially catastrophic pregnancy complication. We report the first case of a heterotopic cervical pregnancy managed in our centre. Selective foetal reduction of the cervical pregnancy with ultra-sonographically guided potassium chloride instillation into the amniotic sac combined with cervical cerclage resulted in the successful preservation of the intrauterine pregnancy and delivery of a live healthy baby at term. This report aims to alert practising clinicians about this rare and fatal pregnancy complication and discuss our experience with its management.

Keywords: Heterotopic cervical pregnancy, selective foetal reduction, assisted reproductive technology, intra-amniotic potassium chloride instillation

Introduction

Heterotopic pregnancy (HTP) is the simultaneous occurrence of intrauterine and extrauterine pregnancy.[1] It is a very rare and potentially life threatening condition with documented incidence rates of between 1in 10,000 50,000 pregnancies.[2] Its incidence is significantly increased following assisted reproductive technique (ART) procedures following which rates of 0.09% to 1.00% have been reported.[3]

A combination of an intrauterine and a cervical heterotopic pregnancy is even rarer and an unusual phenomenon.[2] It is a potentially catastrophic pregnancy as torrential life threatening haemorrhage may complicate the cervical component because of the unsuitability of the cervix for implantation of a fertilized ovum.[4]

A live birth following a cervical heterotopic pregnancy is reported to be a very rare event.[4] We hereby report the first case of a cervical heterotopic pregnancy managed in our centre, with the successful delivery of a live baby at term. To the

best of the knowledge of the authors, this is the first reported case of a heterotopic cervical pregnancy (HCP) in Nigeria with the successful preservation of the intrauterine pregnancy and delivery of a live baby at term. The purpose of this report is to alert practising clinicians about this very rare and potentially fatal complication of pregnancy and discuss our experience with its management.

Case Report

A 34 year old nulliparous lady presented in a private medical facility with secondary infertility of two years duration. There was no previous history of surgery or pelvic infection though she had had two prior first trimester induced abortions. Following evaluation, a diagnosis of male factor infertility was made and she was referred for assisted conception.

She re-presented two years later following a referral from a fertility centre where she had intracytoplasmic sperm injection (ICSI) and embryo transfer (ET) 9 weeks earlier with a trans-vaginal ultrasound (TVS) report that revealed a twin pregnancy with the leading twin around the external cervical os. There was no vaginal bleeding or abdominal pain. Abdominal examination revealed a 14 week pregnant uterus and an enlarged cervix was felt on vaginal examination. She was then referred to the Teaching hospital.

At the Teaching Hospital, previous findings were confirmed. A repeat TVS showed a gravid uterus harbouring two living gestations, one in the uterus (measuring 33 x 24mm) containing an embryo with crown-rump length of 45mm, and the other in the body of the cervix (measuring 43x 39mm) containing an embryo with a crown-rump length of 32.6mm corresponding to an 11 weeks gestation. The cervical gestation was located in the body of the cervix and had symmetrically expanded the cervix, reducing the cervical wall thickness (figure 1).

The patient and her husband were counselled about the possible options of treatment and associated complications and in order to try to conserve the intrauterine pregnancy, they chose selective foetal reduction of the cervical ectopic and an informed consent was obtained.

The patient was scheduled and prepared for ultrasound guided selective foetal reduction with potassium chloride (KCl) as well as cervical cerclage. In theatre, with the patient in the supine position after administration of a subarachnoid block, ultrasonography (USS) was performed to confirm the site and viability of the foetuses. The patient was then placed in the lithotomy position and an examination under anaesthesia performed. Under ultra-sonographic guidance, a Verres needle was passed through the cervical os into the gestational sac in the cevix which was punctured with drainage of 3mls of amniotic fluid. Then about 20mls of KCl was slowly injected into the gestational sac until USS confirmed collapse of the sac and non-viability of the foetus (figure 2). Cervical cerclage was then performed using the McDonald's technique.

Her postoperative period was uneventful. A repeat USS done a week later showed a bulky uterus with a live foetus within the endometrial cavity (crown-rump length of 53.9mm). The gestational sac within the cervical canal had collapsed and was devoid of amniotic fluid. There was no cardiac activity and fragmented placental tissue overlying the distal part of the internal os was seen within the sac (figure 3& 4).The patient was discharged on the seventh postoperative day with no complications.

Subsequently, antenatal care remained uneventful and serial USS examinations done at intervals confirmed a live intrauterine singleton foetus with degenerating foetal matter seen as a heterogeneous mass within the cervix which gradually diminished in size over time until it had been completely reabsorbed. She subsequently had an elective lower segment caesarean section with removal of the cervical stitch at 38 weeks gestation and was delivered of a live female baby weighing 3.3kilograms with good Apgar scores. Her postoperative period was uneventful and she was discharged with her baby on the fifth post-operative day.

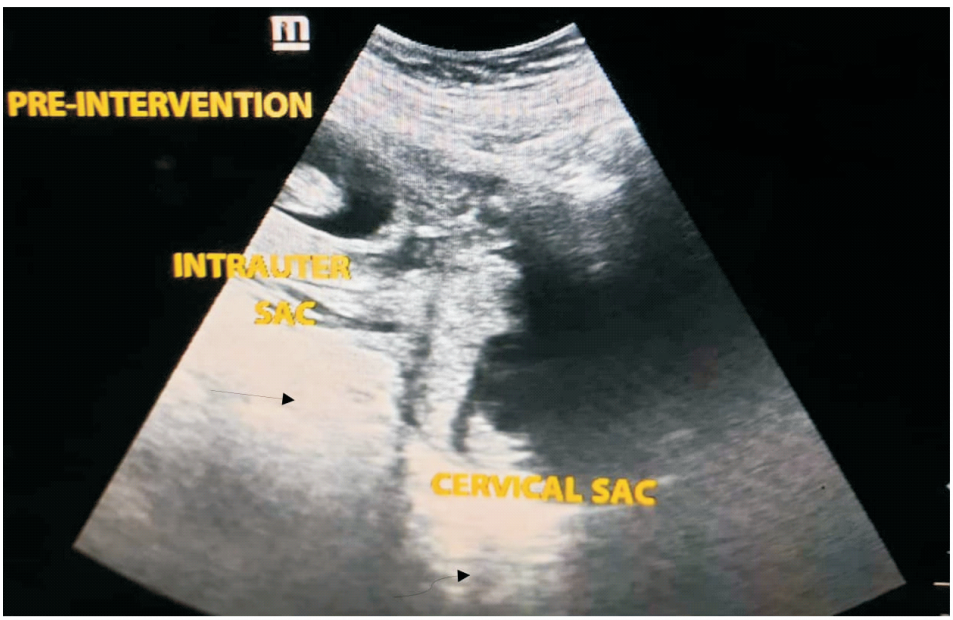


Figure 1 – Longitudinal scanogram showing the viable heterotopic gestations with corresponding placenta. Straight arrow points to the normotopic gestation and posterior placenta. Curved arrow piints to ectopic gestation in the cervical canal with its accompanying placenta.

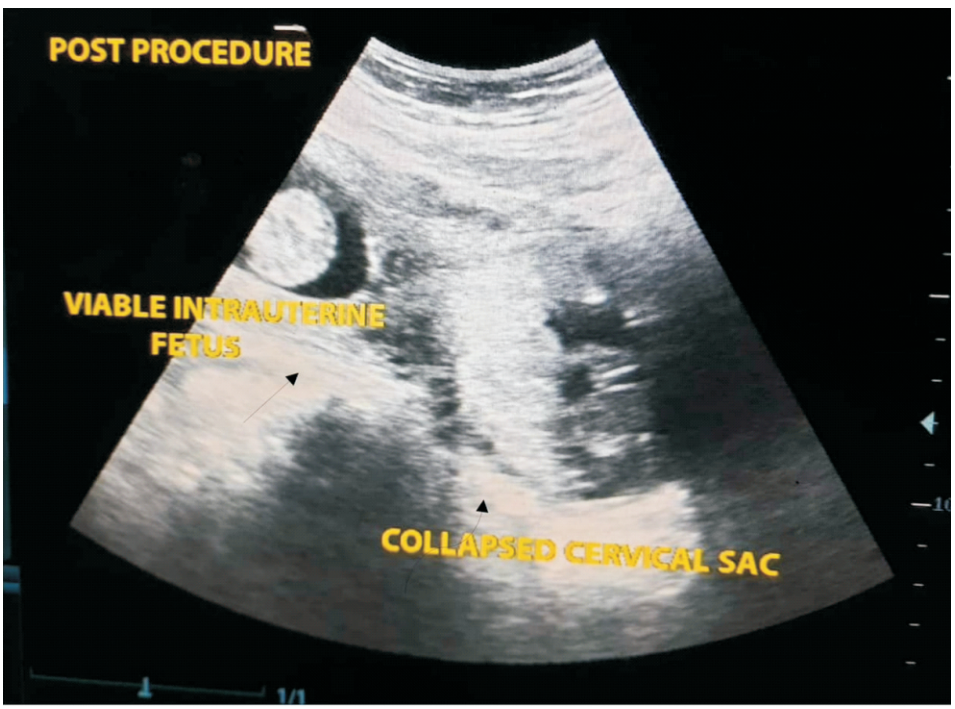
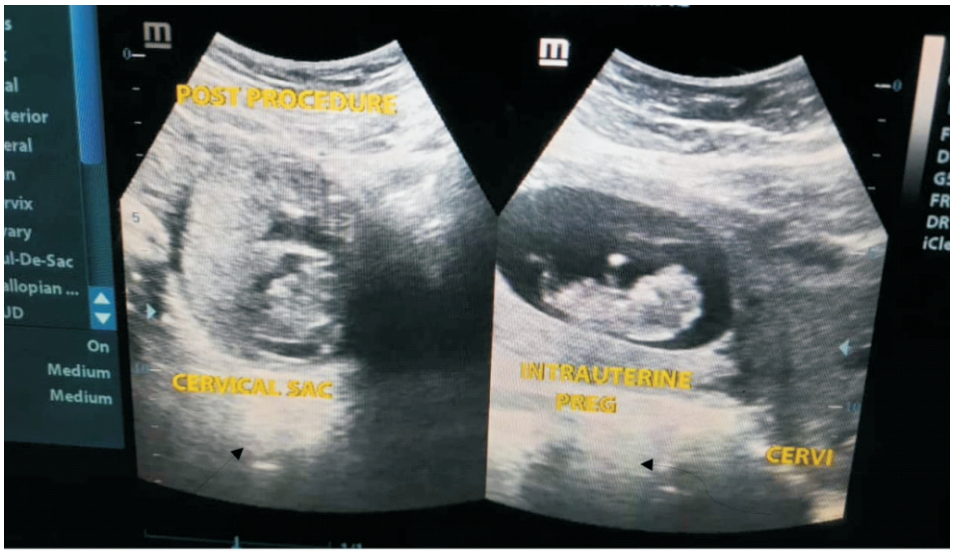


Figure 2 – immediate post procedure echogram shows the viable normotopic intrauterine gestation(straight arrow). The ectopic cervical gestational sac is now collapsed harbouring a disfigured nonviable embryo with sorrounding echogenic debris. Note that its accompanying placenta is still persistent and homogenously echogenic (curved arrow)



Figures 3 and 4 – Immediate post procedure echogram. Figure 3 is the ectopic gestation in the cervical canal with its placenta(straight arrow). This embryo has lost its viability and normal morphology in a partially collapsed sac. Figure 4 refers to normal intrauterine viable gestation withs its placenta(curved arrow).

Discussion

Heterotopic cervical pregnancy is reported to be extremely rare with most cases resulting from ART.[2] It is probably the rarest and most dangerous form of HTP due to the occurrence of serious complications including torrential life threatening haemorrhage from the cervical component which may necessitate emergency hysterectomy.[5]

Due to widespread availability of high resolution TVS and increased expertise in ultrasonography the diagnosis of heterotopic cervical pregnancy can be made early when appropriate management modalities can be instituted in order to successfully preserve the patient's fertility and avoid morbidity and mortality.

Due to its rarity and limited number of cases in the literature, there is no universally accepted treatment modality or established standard protocol for the management of heterotopic cervical pregnancy.[5,6] However, management should individualized based on the clinical presentation and haemodynamic status of the patient, technical availability of the facility, the skill of the managing surgeon and personal characteristics and wishes of the patient and her spouse.[5]

Management can be quite challenging in attempting to preserve the viable intrauterine pregnancy while terminating the cervical one[7] especially in cases such as ours where the patient presented to the Teaching Hospital late in the first trimester and the pregnancy was strongly desired as conception was only ultimately achieved through assisted conception. In addition, in our environment, the burden of infertility is high and very high premium is placed on childbearing.[8]

Conservative approaches to management of heterotopic cervical pregnancy are only possible up to 10 to 12 weeks of pregnancy due to the severe complications that may occur if they are attempted after these gestational ages 6. Generally, the aims of conservative treatment are to protect the coexisting intrauterine pregnancy, preserve the patient's fertility, and minimize complications.[2]

Our patient presented to the Teaching hospital at a gestational age of 11 weeks and had successful selective termination of the cervical ectopic by the instillation of KCl into the amniotic sac of the cervical ectopic under USS guidance combined

with cervical cerclage. KCl is a cardioplegic agent that causes cardiac arrest as the heart stops in asystole.[9] Cervical cerclage was also performed to prevent haemorrhage, and reinforce the internal cervical os. Following the procedure, there were no complications, and the patient was managed successfully till term when she was delivered of a live healthy baby by caesarean section.

Other conservative forms of medical management have been used when attempting to preserve the intrauterine pregnancy. These include injection of methotrexate intracardially or into the amniotic sac under USS guidance, and intra-amniotic injection of hyperosmolar glucose.[10,11] The substance used for selective foetal reduction should have high therapeutic effectiveness and low toxicity to the concurrent pregnancy.[12]

We chose to use KCl and not methotrexate as the agent for terminating the cervical pregnancy because of the risk of systemic absorption of methotrexate and its possible teratogenic effects on the intrauterine foetus. Other surgical forms of conservative management that have been used by other surgeons in a bid to preserve the intrauterine foetus include intra-cervical gestational sac reduction with ring forceps under USS guidance, uterine artery embolization, cervical dilatation and curettage, ultrasound guided aspiration of the cervical pregnancy and hysteroscopic resection of the cervical pregnancy.[4,7,10] However, these methods can be associated with severe vaginal bleeding when attempting to remove the cervical pregnancy because of myometrial involvement. In addition they are also associated with remnants of chorionic tissue due to possible incomplete evacuation which may lead to ascending infection with development of chorioamnionitis, intrauterine foetal death, premature rupture of membranes, severe post-operative bleeding and placenta accreta from chorionic infiltration of the cervix.[2,4,10]

In conclusion, heterotopic cervical pregnancy is an extremely rare and potentially life threatening pregnancy complication whose management can be quite challenging particularly when the intrauterine pregnancy is viable and its preservation is desired. HCP should be sought for and excluded in all successful pregnancies following assisted conception. Ultrasonographically guided KCL instillation into the gestational sac combined with cervical cerclage effectively treated the cervical ectopic in our case and resulted in the successful preservation of the viable intrauterine pregnancy with the delivery of a live healthy baby at term.

References

1. Abasiattai AM, Utuk NM, Ugege W. Spontaneous heterotopic pregnancy with tubal rupture and delivery of a live baby at term: A case report. Nig J Med 2010; 19 :236-238.
2. Kim JW, Park HM, Lee WS, Yoon TK. What is the best treatment of heterotopic cervical pregnancies for a successful pregnancy outcome? Clin Exp Reprod Med 2012; 39: 182-192.
3. Li J, Yang J, Niu G, Fan L, Huang J. Management of heterotopic pregnancy. Medicine 2016; 95:e2750.
4. Deka D, Bahadur A, Singh A, Malhotra N. Successful management of heterotopic pregnancy after foetal reduction using potassium chloride and methotrexate. J Hum Reprod Sci 2012; 5: 57-60.
5. Terra MEF, Giordano LA, Giordano MV, Moreiradesa RA, Compos F, Yadid IM et al. Heterotopic cervical pregnancy after invitro fertilization-case report and literature review. JIBRA Assisted Reprod 2019; 23: 290-296.
6. Hafner T, Ivkosic IE, Serman A, Baurnan R,Ujevic B, Vujisic S et al. Modification of conservative treatment of heterotopic cervical pregnancy by Foley catheter balloon fixation with cervical sutures at the level of the external cervical os: a case report. J Med Case Reports 2010; 4:212.
7. Nitke S, Horowitz E, Farhi J, Krissi H, Shalev J. Combined intrauterine and twin cervical pregnancy managed by a new conservative modality. Fertil Steril 2007; 88:760e1-3.
8. Abasiattai AM, Edemekong II, Bassey EA. Hysterosalpingiographic findings among infertile

women in Uyo, South-Eastern Nigeria. West Afr J Rad 2007: 14: 24-28.
9. Barboza de Oliveira MA, Brandi AC, dos Santos CA, Boldho PHH, Cartex JLL, Braile MD. Modes of induced cardiac arrest: hyperkalemia and hypocalcemia- Literature review. Rev Bras Cir Cardiovasc Sur 2014; 29: 432-436.
10. Faschingbauer F, Mueller A, Voigt F, Beckmann MW, Goeoke TW. Treatment of heterotopic cervical pregnancies. Fertil Steril 2011; 95:1787e9-13.
11. Thangavelu M, Kalkat R. Heterotopic cervical pregnancy. J Clin Gynaecol Obstet 2015; 4: 307-311.
12. Kulhan M, Kulhan NG, Nayki U, Nayki C, Ata N. A rare form of heterotopic pregnancy: cervical pregnancy and intrauterine pregnancy. Arch Med Sci Dis 2017; 2: e110-112.