

Heterotopic pregnancy following intrauterine insemination: Successful management with salpingectomy and continuation of intrauterine pregnancy

Jl Ikechebelu^{1,2}, GU Eleje¹

¹Department of Obstetrics/Gynaecology, Nnamdi Azikiwe University Teaching Hospital, Nnewi and ²Fertility Unit of Life Specialist Hospital Limited, Nnewi Anambra State Nigeria

Abstract

We report the first case of a heterotopic pregnancy (HP) following ovulation induction and intrauterine insemination (IUI) with resultant normal intrauterine pregnancy after salpingectomy. A 41-year-old para 0⁺⁰ that presented with primary infertility due to azoospermia and polycystic ovaries after laparoscopic evaluation. She had induction of ovulation with Clomiphene citrate, gonadotropin stimulation (hCG), and intrauterine insemination using donor sperm. The resulting pregnancy was later diagnosed as heterotopic pregnancy following rupture of the tubal component at 8 weeks' gestation after an initial misdiagnosis as corpus luteum cyst of pregnancy. She had an emergency laparotomy and left salpingectomy, and the intrauterine pregnancy has continued subsequently to 25 weeks of gestation as at 01/04/2011. This report demonstrates that HP may occur after ovulation induction and IUI. The ectopic component could be misdiagnosed as corpus luteum cyst. It is recommended that pregnancies following this procedure be followed up with serial trans-vaginal ultrasound in the first trimester. Presence of corpus luteum cyst of pregnancy in early ultrasound should be an index of suspicious of a possible heterotopic pregnancy. Early diagnosis and prompt intervention is essential to salvage the intrauterine pregnancy and avoid maternal morbidity and mortality.

Key words: Heterotopic pregnancy, intrauterine insemination, salpingectomy, ultrasound

Date of Acceptance: 27-Apr-2011

Introduction

A heterotrophic pregnancy is defined as the presence of a combined intrauterine and ectopic pregnancy.^[1,2] The estimated incidence is between 1/7000 and 1/30 000 pregnancies.^[1] It is also reported to be as high as 1% after the use of assisted reproductive technology, but Clomiphene citrate which increases the rate of twinning, could be associated with a heterotopic pregnancy rate of 1/900, which is much less than using assisted reproductive technology.^[1] Heterotopic pregnancy often poses diagnostic and therapeutic challenges for obstetricians. If this continues without diagnosis, a life-threatening situation

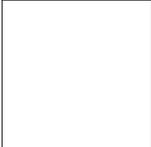
may occur even when surgical intervention with laparotomy is performed.

In one systematic review, it was observed that compared with timed intercourse, intrauterine insemination (IUI) resulted in increased pregnancy rates, both in natural cycles and stimulated cycles.^[3] If any form of ovulation induction has been used it is also quite common to use a single human chorionic gonadotrophin (hCG) injection approximately 36 h prior to the insemination to ensure optimal timing with ovulation.^[1,4]

Address for correspondence:

Prof. Joseph I Ikechebelu,
P.O.Box 244 Nnewi, Anambra State Nigeria
E-mail: ji.ikechebelu@unizik.edu.ng

Access this article online

Quick Response Code: 	Website: www.njcponline.com
	DOI: ***
	PMID:

We present a case of heterotopic pregnancy following IUI in our fertility practice in Nnewi, Nigeria.

Case Report

Mrs. AI was a 41-year-old Gravida 1 Para 0⁺⁰. She was referred to our facility due to primary infertility of 5 years duration. There was no previous history of tubal surgery, pelvic infections, termination of pregnancy or use of hormonal contraceptives. Laparoscopy and dye test revealed bilateral patent tubes and bilateral polycystic ovaries. The uterus was normal and there were no pelvic adhesions. There were no uterine fibroids. Seminalysis showed azoospermia and scrotal ultrasound revealed low volume atrophic testicles. The couple were counseled on the diagnosis and superovulation and intrauterine insemination using donor sperm were recommended.

She received one course of 100 mg clomiphene citrate daily for 5 days from the second day of her cycle (12/10/2010) followed by serial trans-vaginal ultrasound for follicular tracking from day 10 till day 13 of cycle. Day 13 ultrasound revealed multiple mature follicles, 3 per each ovary. She received 10 000 IU of hCG on day 13 and, subsequently, had intrauterine insemination on 27/10/2010 using donor sperm 36 h after hCG injection. The outcome was satisfactory. The urine pregnancy test was positive four weeks after IUI.

Trans-vaginal ultrasound was also done and revealed single fetal pole with visible fetal heart activity and gestational sac diameter of 19 mm at a gestational age of 6 weeks and 1 day. Concurrently, the left adnexum had a spongy mass, which was regarded as a corpus luteum cyst. She was counselled and reassured of the findings. She was allowed home and subsequently was placed on pregnancy support using intramuscular progesterone caproate (primolut) depot 250 mg weekly.

However, one week later, she started having vaginal spotting, and no abdominal pains. This was managed conservatively with bed rest. The symptoms persisted after 1 week and repeat trans-vaginal ultrasound on 7/12/2010 (at 8 weeks of gestation) revealed simultaneous left tubal ectopic pregnancy and a viable intrauterine pregnancy with moderate haemoperitoneum. A diagnosis of heterotopic pregnancy with rupture of the tubal pregnancy was made. She was counselled for emergency laparotomy and the couple gave consent.

At laparotomy, a significant haemoperitoneum of about 500 ml was found. After clearing the clots from the pelvis, a mass within the left fallopian tube was identified, consistent with an ectopic pregnancy. The mass was approximately 6 × 3 cm, with a linear rupture at the ampullary region. A left salpingectomy was then performed with minimal

handling of the uterus. The patient recovered well and was discharged home on postoperative day 8 and the intrauterine gestation is still viable. Histopathologic examination of the left fallopian tube and its contents revealed chorionic villi, confirming the diagnosis of a tubal pregnancy.

Further abdominal ultrasound on 15/01/2011 (six weeks after the laparotomy) revealed a viable intrauterine singleton fetus with crown rump length of 89 mm with a gestational age of 14 weeks five days. She is receiving antenatal care and the pregnancy as of 01/04/2011 was 25 weeks of gestation.

Discussion

Heterotopic pregnancy has been traditionally considered a rare event. However, with the use of assisted reproductive technology, the incidence of heterotopic pregnancies is increasing. Diagnosing a heterotopic pregnancy can be challenging as in this case report. However, the diagnosis is imperative so that treatment can be initiated in a timely manner.

A recent case series demonstrated that patients with heterotopic pregnancies are more likely to have tubal rupture and present in hypovolaemic shock.^[5] In this patient, the diagnosis was delayed because of a confusing clinical presentation and an initial ultrasound regarding the left adnexal mass as corpus luteum cyst. The patient was not acutely ill despite a significant haemoperitoneum from tubal rupture and had minimal vaginal bleeding. Ectopic and heterotopic pregnancies are frequently diagnosed between the early and mid first trimester^[6] as was the case in this patient.

As was seen in this case, patients should also be counselled about the possibility of ectopic pregnancy following super-ovulation and IUI. Nevertheless, most clinics would offer an early ultrasound scan in patients who have had a positive pregnancy test, at between 6 and 7 weeks of gestation^[3] as was done in our patient.

Additionally, in cases where there is either azoospermia or necrozoospermia surgical sperm retrieval can be performed to obtain sperm directly from either the epididymis (microepididymal sperm aspiration – MESA) or directly from the testis (testicular sperm extraction – TESE) or (percutaneous epididymal sperm aspiration – PESA).^[2,3] However, this does not apply to our patient's husband who had testicular atrophy and no sperm production. Donor insemination was resorted to. IUI is relatively a simple technique that is cost-effective and can be offered by both secondary and tertiary fertility centres. It is not as invasive as IVF and allows fertilization to occur within the fallopian tubes and therefore is generally acceptable to most religious

groups even as an alternative to having sexual intercourse with other men.^[2,3,7]

Since heterotopic pregnancy is a rare event, no standardized management recommendations currently exist. Mrs AI was treated with salpingectomy following an emergency laparotomy. However, other treatment options have been described. Several case reports and series have described successful treatment of a heterotopic pregnancy with trans-vaginal embryo aspiration of the ectopic under ultrasound guidance followed by local instillation of methotrexate or potassium chloride.^[8-10] Surgical treatment remains the most common therapy in our environment. As in our case, laparotomy is frequently used when a patient is haemodynamically unstable and or when large haemoperitoneum is suspected. However, in hemodynamically stable patients, laparoscopy can be successfully and safely carried out.^[5,11,12]

Conclusion

This report has demonstrated that heterotopic pregnancy may occur following ovulation induction and IUI. It is recommended that early first trimester trans-vaginal ultrasound be performed for pregnancies resulting from this procedure and coexisting corpus luteum cyst with intrauterine gestation should raise an index of suspicion for possible heterotopic pregnancy. This case demonstrates that early diagnosis is essential in order to salvage the intrauterine pregnancy and avoid maternal morbidity and mortality.

Acknowledgments

We thank Dr. R.C. Okwuosa of Ajefo Okwuosa Memorial Hospital Ogidi for providing us with the ultrasound report and the operative findings on this patient.

References

1. Honarbakhsh A, Khoori E, Mousavi S. Heterotopic pregnancy following ovulation induction by Clomiphene and a healthy live birth: A case report. *J Med Case Reports* 2008;2:390.
2. Edmonds DK. Assisted Reproduction. In: Edmonds DK, editor. *Dewhurst's Textbook of Obstetrics and Gynaecology*. 7th ed. London: Blackwell Science; 2007. p. 369-76.
3. Cohlen BJ, Vandekerckhove P, te Velde ER, Habbema JD. Timed intercourse versus intra-uterine insemination with or without ovarian hyperstimulation for subfertility in men. *Cochrane Database Syst Rev* 2000;(2): CD000360.
4. Drinville JS, Memarzadeh S. Benign Disorders of the Uterine Corpus. In: De Cherney AH, Nathan L, editors. *Current Obstetrics and Gynaecologic Diagnosis and Treatment*. 10th ed. New York: Lange McGraw Hill; 2007. p. 639-53.
5. Soriano D, Shrim A, Seidman DS, Goldenberg M, Mashiach S, Oelsner G. Diagnosis and treatment of heterotopic pregnancy compared with ectopic pregnancy. *J Am Assoc Gynecol Laparosc* 2002;9:352-8.
6. Childs AJ, Royek AB, Leigh TB, Gallup PG. Triplet heterotopic pregnancy after gonadotropin stimulation and intrauterine insemination diagnosed at laparoscopy: A case report. *South Med J* 2005;98:833-5
7. Ikechbelu JI, Okeke CA. Intrauterine insemination with controlled ovarian stimulation for treatment of infertility: An audit of a new IUI programme in a low resource setting. *J Obstet Gynaecol* 2007;27:61-4.
8. Oyawoye S, Chander B, Pavlovic B, Hunter J, Gadir AA. Heterotopic pregnancy: Successful management with aspiration of cornual/interstitial gestational sac and instillation of small dose of methotrexate. *Fetal Diagn Ther* 2003; 18:1-4.
9. Porreco R, Burke MS, Parker DW. Selective embryocide in the nonsurgical management of combined intrauterine-extrauterine pregnancy. *Obstet Gynecol* 1990;75:498-501.
10. Wright A, Kowalczyk CL, Quintero R, Leach RE. Selective embryo reduction in a heterotopic pregnancy using potassium chloride injection resulting in a hematosalpinx. *Fertil Steril* 1996;66:1028-30.
11. Chang Y, Lee JN, Yang CH, Hsu SC, Tsai EM. An unexpected quadruplet heterotopic pregnancy after bilateral salpingectomy and replacement of three embryos. *Fertil Steril* 2003;80:218-220.
12. Hoopmann M, Wilhelm L, Possover M, Nawroth F. Heterotopic triplet pregnancy with bilateral tubal and intrauterine pregnancy after IVF. *Reprod Biomed Online* 2003;6:345-8.

How to cite this article: ???

Source of Support: Nil, **Conflict of Interest:** None declared.