# Combined Tubal And Ovarian Ectopic Pregnancies In One Patient After In Vitro Fertilization

İn Vitro Fertilizasyondan Sonra Aynı Hastada Kombine Tübal ve Ovaryan Ektopik Gebelikler

## Yusuf Üstün, Yaprak Engin Üstün, Hale Kırımlıoğlu, İlgın Türkçüoğlu

İnönü Üniversitesi Tıp Fakültesi Turgut Özal Tıp Merkezi Kadın Hastalıkları ve Doğum ABD Malatya

A case of twin ectopic ovarian-tubal pregnancy located at the same side after an intracytoplasmic sperm injection-embryo transfer cycle is reported. The case was refered to our clinic for suspected ectopic pregnancy 27 days later the transfer of 3 embryos, with lower abdominal pain. Transvaginal ultrasound scan revealed 17 mm gestational sac with a yolk sac inside and a 15 mm echo-complex ovarian mass at the left adnexa. At laparotomy a ruptured 2 cm ovarian hemorrhagic mass and an unruptured 2 cm fimbrial ectopic pregnancy was found at the left adnexa. A left partial salphengectomy and wedge resection of the left ovary was performed. The histopathology showed the presence of chorionic villi both in the ovarian tissue and the left fallopian tube.

#### Key Words : IVF, Ectopic Pregnancy, Tubal, Ovarian

İntrasitoplazmik sperm enjeksiyonu-embryo transferi siklusundan sonra aynı tarafta gelişen ovaryan-tubal ikiz ektopik gebelik olgusu bildirilmiştir. Vaka, alt kadranlarda karın ağrısı şikayeti ile, 3 embryo transferinden 27 gün sonra ektopik gebelik şüphesi ile kliniğimize refere edildi. Transvajinal ultrasonografide sol adneksiyal alanda içerisinde yolk kesesi bulunan 17 mm gestasyonel kese ve 15 mm eko-kompleks ovaryan kitle tesbit edildi. Laparotomide, sol adnekste 2 cm rüptüre hemorajik ovaryan kitle ve 2 cm intakt fimbrial ektopik gebelik ile uyumlu kitle görüldü. Sol parsiyel salfenjektomi ve sol overe wedge rezeksiyon yapıldı. Histopatoloji incelemede hem over dokusunda hem de fallopi tüpünde koryonik villusların varlığı gösterildi.

Anahtar Sözcükler: IVF, Ektopik Gebelik, Tübal, Ovaryan

- Ovarian pregnancy is a rare event, with estimated frequency ranging from 1 in 2100 to 1 in 7000 pregnancies (1). Assisted reproductive technologies increased incidence of ectopic pregnancy, however primary ovarian pregnancy is still rare (2). Following in vitro fertilization – embryo transfer (IVF – ET) cycles, the overall prevalence of ovarian pregnancy has been estimated to be 0.3%, representing 6% of all ectopic pregnancies (3).
- The first published case of twin ectopic pregnancy was unilateral tubal pregnancy reported by De Ott in 1891 (4). Since then, about 250 twin ectopic pregnancies have been reported (5-9). Primary ovarian pregnancy as a component of twin ectopic pregnancy is very rare.

We reported a case of twin ectopic ovarian – tubal pregnancy located at the same side after an intracytoplasmic sperm injection – embryo transfer (ICSI – ET) cycle.

## **Case Report**

A 32 year-old female, gravida 1, para 0, was referred to our hospital for suspected ectopic pregnancy, with a lower abdominal pain. She didn't have any pregnancy for 13 years due to oligoastenozoospermia. She had ICSI – ET 27 days ago and 3 embryos were transferred under ultrasound guidance. She denied any past history of pelvic inflammatory disease, intrauterine device use and previous surgery.

Physical examination revealed tenderness in the left pelvic region. Her systolic

#### Received: 12.08.2009 · Accepted: 05.10.2009

Corresponding author

 Dr. Ilgın Türkçüoğlu

 İnönü Üniversitesi Tıp Fakültesi Turgut Özal Tıp Merkezi Kadın

 Hastalıkları ve Doğum ABD Malatya

 Phone
 : +90 (422) 341 06 60 / 47 05

 Fax
 : +90 (422) 341 07 28

 E-mail Address
 : dr.ilgin@yahoo.com

blood pressure was 90 mmHg, diastolic blood pressure was 60 mmHg and pulse rate was 120 beats/min. Serum  $\beta$ -hCG level was 26723 mIU/ml and hemoglobin level was 12.6 g/dl.

- Transvaginal ultrasound scan revealed an empty uterine cavity with an endometrial thickness of 11.9 mm. At the left adnexa a 17 mm gestational sac with a yolk sac inside and a 15 mm echocomplex ovarian mass were found. The right tubo¬ovarian region was normal. Free fluid was seen in the pouch of Douglas. The sonographic findings and the serum  $\beta$ -hCG level suggested a ruptured ectopic pregnancy.
- She underwent emergent laparotomy due to hemorrhagic shock. The abdomen was opened through a pfannenstiel incision. Exploration revealed 500 ml of blood and clots in the abdominal cavity. The uterus, right ovary and right fallopian tube appeared normal. The left ovary was enlarged with a 2 cm hemorrhagic mass appearance, which was the source of bleeding. There was a fimbrial ectopic pregnancy of 2 cm in diameter in the left tube which was intact, though blood was trickling from the fimbrial edge of the tube. A left partial salphengectomy and wedge resection of the left ovary was performed. The histopathology of the specimen showed the presence of chorionic villi both in the ovarian tissue and the left fallopian tube supporting the diagnosis of twin ectopic ovarian - tubal pregnancy (Figure 1).
- Post-operatively the patient received antibiotics. Serum  $\beta$ -hCG level was followed up and the values showed a progressive decline confirming the efficiency of the treatment. After the stabilization of the patient and the detection of gradual decline in the  $\beta$ -hCG levels the patient was discharged from the hospital and followed by  $\beta$ -hCG level until it was under 1 mIU/ml.

## Discussion

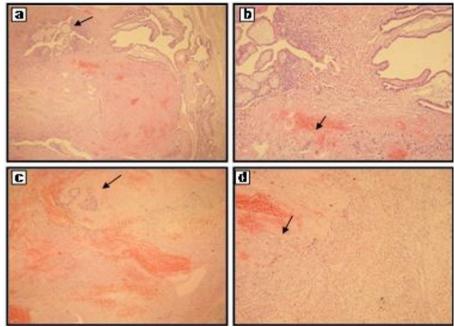
In cases of assisted conception using IVF -

ET, the complication of ectopic pregnancy is relatively common, occurring in 1-3 % of these pregnancies (10). Twin ectopic pregnancy rate is much less than single ectopic pregnancy. The most common form is twin tubal gestations (7, 8). Ovarian (11)., interstitial (12). and abdominal (6). twin pregnancies have also been reported. To our knowledge, this is the first case of twin ectopic pregnancy consisting of ectopic ovarian and tubal pregnancy at the same side after an ICSI – ET cycle.

- Factors predisposing for ectopic pregnancy are tubal damage after pelvic inflammatory disease, endometriosis or tubal surgery, previous ectopic pregnancies, progesterone intrauterine device and exposure to diethylstilbestrol in utero. Also there is a strong association between ovarian pregnancies and current use of intrauterine device (IUD) (13). These predisposing factors were not present in our case.
- In assisted reproduction cycles utilizing IVF /ICSI and embryo transfer there are some theoretical risk factors for ectopic implantation; including reduction in tubal contractility as a result of high progesterone levels from multiple corpus lutea, ovarian hypervascu-

larity after hyperstimulation and egg retrieval, excessive endometrial cavity distention with media during embryo transfer, deep fundal embryo transfer, high number of the transferred embryos, and transfer of blastocyst (3, 14-18). It has been hypothesized that, even correctly transferred embryos can migrate into fallopian tubes, due to retrograde action of uterine secretions and /or uterine contractions(19). In the case we described the exact mechanism of ovarian and tubal pregnancy after ICSI was not clear since there was no predisposing factors. The most probable mechanism is reverse migration of two separate embryos toward the fallopian tube and implantation in the ovary and tuba at the same side.

Demonstration of a live embryo within a gestational sac outside the uterus is the gold standard for the sonographic diagnosis of ectopic pregnancy. However yolk sac and/or embryo is seen relatively infrequent both in ovarian and tubal pregnancies (20-22). Correct preoperative diagnosis of ovarian pregnancy is difficult, being confused with corpus luteal cysts (23). In review of 25 ovarian pregnancies, the correct diagnosis was made surgically in only 28% of cases and an embryo was



**Figure 1:** Tubal (1a and 1b) and ovarian (1c and 1d) ectopic pregnancies were shown in the figure. Both chorion villi (arrows in figure 1a and 1c) and trophoblastic cells (arrows in figure 1b and 1d) were seen.

Combined Tubal And Ovarian Ectopic Pregnancies In One Patient After In Vitro Fertilization

identified in only 12% of cases (24). In a standard IVF-ET cycle diagnosis of ovarian pregnancy is harder since the initial sonographic picture might be obscured by multiple corpora lutea cysts after hyperstimulation and egg retrieval (25).

Surgery is the gold standard for the treatment of ovarian pregnancies. Ovarian preserving surgery; either cystectomy or wedge resection done by laparoscopy or laparotomy is the preferred treatment option(26). Although laparoscopic approach is the first choice especially in the early diagnosed ca-

### REFERENCES

- Hage PS, Arnouk IF, Zarou DM, Kim BH, Wehbeh HA. Laparoscopic management of ovarian ectopic pregnancy. J Am Assoc Gynecol Laparosc. 1994 May;1(3):283-5. Review.
- Abusheikha N, Salha O, Brinsden P. Extrauterine pregnancy following assisted conception treatment. Hum Reprod Update. 2000 Jan-Feb;6(1):80-92. Review.
- Marcus SF, Brinsden PR. Primary ovarian pregnancy after in vitro fertilization and embryo transfer: report of seven cases. Fertil Steril. 1993 Jul;60(1):167-9.
- 4. De Ott. A case of unilateral tubal twin gestation. Ann Gynecol Obstet 1891;36:304.
- Ansari SM, Nessa L, Saha SK. Twin ectopic pregnancy 10 years after permanent sterilization. Australas Radiol. 2000 Feb;44(1):107-8.
- Deshpande N, Mathers A, Acharya U. Broad ligament twin pregnancy following in-vitro fertilization. Hum Reprod. 1999 Mar;14(3):852-4.
- Hanchate V, Garg A, Sheth R, Rao J, Jadhav PJ, Karayil D. Transvaginal sonographic diagnosis of live monochorionic twin ectopic pregnancy. J Clin Ultrasound. 2002 Jan;30(1):52-6.
- Parker J, Hewson AD, Calder-Mason T, Lai J. Transvaginal ultrasound diagnosis of a live twin tubal ectopic pregnancy. Australas Radiol. 1999 Feb;43(1):95-7.

ses, in hemodynamically unstable cases with a ruptured ectopic pregnancy, laparotomy is mostly performed. In hemodynamically unstable patients we prefer laparotomy in our clinic. Ovarian wedge resection and unilateral salphengectomy was done in this case in order to preserve future fertility. Methotrexate has become an increasingly popular treatment for ectopic pregnancies (27). Treatment with methotrexate may be particularly helpful in preserving the ovary in patients with a preoperative diagnosis of ovarian pregnancy.

- Marret H, Hamamah S, Alonso AM, Pierre F. Case report and review of the literature: primary twin ovarian pregnancy. Hum Reprod. 1997 Aug;12(8):1813-5. Review.
- 10.Karande VC, Flood JT, Heard N, Veeck L, Muasher SJ. Analysis of ectopic pregnancies resulting from in-vitro fertilization and embryo transfer. Hum Reprod. 1991 Mar; 6(3): 446-9.
- 11.Einenkel J, Baier D, Horn LC, Alexander H. Laparoscopic therapy of an intact primary ovarian pregnancy with ovarian hyperstimulation syndrome: case report. Hum Reprod. 2000 Sep;15(9):2037-40.
- 12.Ophir E, Singer-Jordan J, Oettinger M, Odeh M, Tendler R, Feldman Y, Fait V, Bornstein J. Uterine artery embolization for management of interstitial twin ectopic pregnancy: case report. Hum Reprod. 2004 Aug;19(8):1774-7. Epub 2004 Jun 24.
- 13.Sandvei R, Ulstein M. History and findings in ectopic pregnancies in women with and without an IUD. Contracept Deliv Syst. 1980 Apr;1(2):131-8.
- 14.Oliveira FG, Abdelmassih V, Costa AL, Balmaceda JP, Abdelmassih S, Abdelmassih R. Rare association of ovarian implantation site for patients with heterotopic and with primary ectopic pregnancies after ICSI and blastocyst transfer. Hum Reprod. 2001 Oct;16(10):2227-9.

## Conclusion

IVF-ET increases the incidence of ectopic pregnancy especially in unpredictable locations. Even without known ectopic pregnancy risk factor, in women submitted to IVF-ET, it's mandatory to perform an early  $\beta$ -hCG monitoring and transvaginal ultrasonography in order to detect ectopic pregnancy at an early stage for a chance of possible conservative treatment.

- 15.Gaudoin MR, Coulter KL, Robins AM, Verghese A, Hanretty KP. Is the incidence of ovarian ectopic pregnancy increasing? Eur J Obstet Gynecol Reprod Biol. 1996 Dec 27;70(2):141-3.
- 16.Tal J, Haddad S, Gordon N, Timor-Tritsch I. Heterotopic pregnancy after ovulation induction and assisted reproductive technologies: a literature review from 1971 to 1993. Fertil Steril. 1996 Jul;66(1):1-12.
- 17.Atabekoglu CS, Berker B, Dunder I. Ovarian ectopic pregnancy after intracytoplasmic sperm injection. Eur J Obstet Gynecol Reprod Biol. 2004 Jan 15;112(1):104-6.
- 18.Yovich JL, Turner SR, Murphy AJ. Embryo transfer technique as a cause of ectopic pregnancies in in vitro fertilization. Fertil Steril. 1985 Sep;44(3):318-21.
- 19.Hemmings R, Biljan MM, Dean N, Tan SL. An ectopic pregnancy masked by follicular initiation of gonadotropin-releasing hormone agonist for pituitary desensitization prior to in vitro fertilization. J Assist Reprod Genet. 1998 Mar;15(3):161-3.
- 20.de Crespigny LC. Demonstration of ectopic pregnancy by transvaginal ultrasound. Br J Obstet Gynaecol. 1988 Dec;95(12):1253-6.
- 21.Stabile I, Campbell S, Grudzinskas JG.Can ultrasound reliably diagnose ectopic pregnancy? Br J Obstet Gynaecol. 1988 Dec;95(12):1247-52.
- 22.Comstock C, Huston K, Lee W. The ultrasonographic appearance of ovarian ec-

topic pregnancies. Obstet Gynecol. 2005 Jan;105(1):42-5.

- 23.Raziel A, Golan A, Pansky M, Ron-El R, Bukovsky I, Caspi E. Ovarian pregnancy: a report of twenty cases in one institution. Am J Obstet Gynecol. 1990 Oct;163(4 Pt 1):1182-5.
- 24.Hallatt JG. Primary ovarian pregnancy: a report of twenty-five cases. Am J Obstet Gynecol. 1982 May 1;143(1):55-60.
- 25.Ranieri DM, Sturdy J, Marchant S, Kinis A, Serhal P. Ovarian heterotopic pregnancy after IVF and contralateral tubal ectopic pregnancy after GIFT. Acta Eur Fertil. 1992 Sep-Oct;23(5):243-5
- 26.Seinera P, Di Gregorio A, Arisio R, Decko A, Crana F. Ovarian pregnancy and operative laparoscopy: report of eight cases. Hum Reprod. 1997 Mar;12(3):608-10
- 27.Annunziata N, Malignino E, Zarcone R. Ovarian pregnancies treated with methotrexate. Panminerva Med. 1996 Sep;38(3):190-2