

A case of heterotopic pregnancy in a spontaneous cycle

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Abstract

Objective: Heterotopic pregnancy is a condition in which extrauterine and intrauterine pregnancy coincides. It is encountered very rarely especially in the absence of risk factors. The incidence of heterotopic pregnancy has increased in recent years due to the developments in assisted reproductive technologies (ART). In this case report, we aimed to emphasize the early diagnosis and proper management of heterotopic pregnancies with its mortality and morbidity rates and importance for future fertility.

Case: Seven-weeks pregnant women by the date of her last menstrual period had undergone dilatation and curettage (D&C) in another medical center upon the diagnosis of a missed abortion. She applied to the same center after the procedure with the complaint of groin pain and was referred to our tertiary center because of the plateaued β -hCG levels during her follow-up. Her gynecologic and ultrasonographic examination in our clinic suggested an extrauterine pregnancy and the patient underwent a laparoscopy. A tubal ectopic pregnancy was detected at the right tube and a linear salpingotomy was performed. Heterotopic pregnancy diagnosis was confirmed with the pathological examination reports of the samples obtained in D&C and laparoscopy.

Conclusion: The heterotopic pregnancy diagnosis should be kept in mind in the differential diagnosis of women who applied with the complaint of abdominal pain in the first trimester of pregnancy especially in pregnancies occurring after ART and should be managed appropriately on time.

Key words: Heterotopic pregnancy, ART pregnancies, linear salpingotomy.

Spontan siklуста heterotopik gebelik olgusu

Amaç: Heterotopik gebelik; intrauterin ve ekstrauterin gebeliğin birlikte bulunduğu, özellikle risk faktörleri bulunmadığında oldukça nadir rastlanılan bir durumdur. Son yıllarda yardımcı üreme tekniklerindeki gelişmeler sonucunda heterotopik gebelik insidansında artış meydana gelmiştir. Bu olgu sunumunda heterotopik gebeliklerin erken tanı ve uygun yaklaşımla tedavileri sonucu mortalite, morbidite oranlarını ve gelecekteki fertilité açısından önemini vurgulamayı hedefledik.

Olgu: Son adet tarihine göre 7 haftalık gebe olan hastanın antenatal takiplerinde, missed abortus saptanması üzerine bir başka sağlık merkezinde dilatasyon küretaj işlemi uygulanmıştır. İşlem sonrası kasık ağrısı şikayetiyle yine aynı merkeze başvuran hastanın yapılan β -hCG takiplerinin plato yapması üzerine kliniğimize sevk edilmiştir. Kliniğimizde olgunun jinekolojik ve ultrasonografik muayenesinde sağ tubal ektopik gebelik saptanmış olup laparoskopik sağ lineer salpingotomi operasyonu uygulanmıştır. Heterotopik gebelik tanısı küretaj ve laparoskopi de alınan materyallerin patolojik incelemeleri sonuçları ile doğrulanmıştır.

Sonuç: Özellikle üremeye yardımcı tedavi teknikleri sonucu oluşan gebelikler olmak üzere, tüm gebeliklerin ilk trimesterinde, karın ağrısı şikayeti ile başvuran olguların ayırıcı tanısında heterotopik gebelik akılda tutulmalı ve zamanında uygun şekilde tedavi edilmelidir.

Anahtar sözcükler: Heterotopik gebelik, üremeye yardımcı tedavi teknikleri gebelikleri, lineer salpingotomi.

Introduction

Presence of intrauterine and extrauterine pregnancies at the same time is called as heterotopic pregnancy. Although extrauterine pregnancy is usually on tubes, it may be rarely ovarian, cervical, cornual, old cesarean

scar or abdominal located.^[1] It was first defined by Duverney in 1708 as the autopsy finding in a patient who died due to ruptured ectopic pregnancy and also had intrauterine pregnancy.^[2] Its reported prevalence is 0.08% in normal conception.^[3] However, this rate

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increases up to 1 in 100 pregnancies in infertile women treated by assisted reproductive technologies (ART).^[4]

Heterotopic pregnancy which may cause inferior pelvic and groin pains or acute abdominal indications especially in first trimester also may cause severe maternal morbidity and even mortality if it is diagnosed lately.^[5] Therefore, although it is not a common pathology, heterotopic pregnancy possibility should not be ignored in the differential diagnosis of patients who applied with the complaint of acute abdominal pain in the first trimester. By reviewing the literature, we aimed in our study to present a heterotopic pregnancy case which developed after natural conception and that we established its early diagnosis and treated accordingly.

Case Report

Thirty-two-year-old (gravida 2, parity 0) patient who applied to a secondary healthcare organization upon the complaint of menstrual delay after natural cycle was diagnosed as pregnant for 7 weeks and missed abortion. Dilatation and curettage (D&C) was applied to the patient upon her request in the same center. The patient applied to the same health center 4 days later with the complaint of groin pain and minimal right adnexal sensitivity was found on gynecologic examination, the uterine cavity was found normal, endometrial echo was found thin and both adnexal regions were found natural. The pathology result of dilatation and

curettage material was reported as “chorial villi surrounded by syncytial cells, and endometrium including desidual cells”; and the case was taken into follow-up when β -hCG values of the case was found as 3,200 IU/L before curettage and as 3,900 IU/L after curettage. When β -hCG values of the case was found as 4,900 IU/L four days later, the patient was referred to our clinic with the diagnosis of “gestational trophoblastic disease”.

In the pelvic examination of the patient performed in our clinic, right lower quadrant sensitivity was detected and β -hCG value was reported as 4,500 IU/L. Gestational trophoblastic disease was considered as pre-diagnosis and then remaining placenta pieces and heterotopic pregnancy were considered. In the transvaginal ultrasonographic (TV USG) examination, it was observed that uterus was in normal sizes, endometrial echo was thin and regular, left adnexa was natural and there was mass lesion measuring 15x20 mm on right adnexal canal (**Figure 1a**) which was consistent with ectopic gestational focus; so the diagnosis was interpreted as heterotopic pregnancy.

Treatment alternatives were suggested to the patient and surgery was decided since she did not accept methotrexate treatment. Right tubal ampullary ectopic pregnancy focus was focused in the laparoscopy (**Figure 1b**), right linear salpingotomy was applied to the case and she was discharged with full recovery on first post-operative day. Our diagnosis was confirmed when the material taken by laparoscopy was reported as “tubal

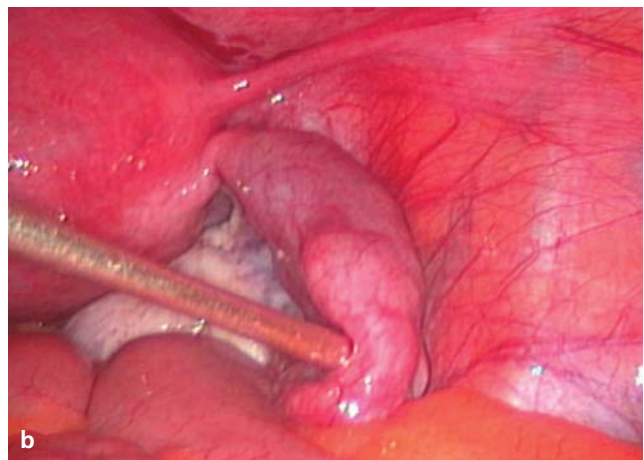
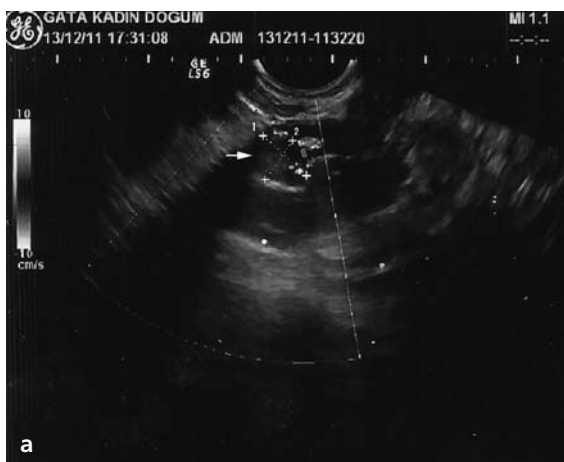


Figure 1. (a) Ultrasonography and (b) surgery images of the case.

ectopic” and “chorionic villus and desidual cells” were confirmed in the pathological examination of paraffin block preparations asked from external centers.

Discussion

Heterotopic pregnancy incidence was reported as 1/30,000 in 1948 by the study of Devoe and Pratt. However, the classical incidence which is known as 1/30,000 in the literature has been revised as 1/3889 in recent analyses and it is anticipated that this rate may increase up to 1/100 in the cycles of assisted reproductive technologies.^[5,6]

The risk factors increasing ectopic pregnancy incidence, which are previous pelvic inflammatory disease (PID), intrauterine device (IUD), assisted reproductive technology, endometriosis, previous abdominal surgery, tubal surgery and sexually transmittable diseases, are also valid for heterotopic pregnancies.^[7] These risk factors can be categorized within two groups for heterotopic pregnancies: (1) while assisted reproductive technologies increases the risk associated with the number of transferred embryo (if more than one), and (2) other risk factors are associated with tubal damage.^[1]

Although extrauterine gestational focus is generally one in heterotopic pregnancy, there are also heterotopic triplet pregnancies in the literature defined as two gestational sacs in one tube and one gestational sac in each tube, and reported after natural conception or assisted reproductive technology (ART) practices.^[8,9]

First symptoms of the heterotopic pregnancy are abdominal pain and vaginal bleeding as in ectopic pregnancy. However, when a patient with such symptoms also has intrauterine pregnancy, it is usually ignored that there may be an accompanying ectopic pregnancy and symptoms are most likely associated with normal or pathological intrauterine pregnancy.^[1] Early diagnoses of these pregnancies are of vital importance in terms of mortality, morbidity and future fertility of patient. However, in case of a normal intrauterine pregnancy, a suspicious lesion ultrasonographically seen on adnexal region can be interpreted as hemorrhagic corpus luteum and this may delay the diagnosis.^[10] In these cases, maternal mortality is reported as 1% and mortality rate of intrauterine fetus is reported as 45-65%.^[11]

Management of heterotopic pregnancy should be customized according to case. β -hCG and progesterone

levels are significant in diagnosis and follow-up as well as determination of pregnancy ultrasonographically. Serial progesterone measurements may determine bad prognosis of pregnancy. However, serial β -hCG follow-ups do not help due to accompanying intrauterine pregnancy. The most significant method for the diagnosis of heterotopic pregnancy is high-resolution TV USG. In high risk patients and especially ART practices, both for intrauterine pregnancy diagnosis and differential diagnosis of ectopic and heterotopic pregnancies after embryo transfer 4-6 weeks later, it is recommended to perform routine ultrasonographic evaluation.^[12]

Although it is reported that only 10% of previous series of heterotopic pregnancy cases which are hardly diagnosed can be diagnosed during preoperative period, ultrasonography sensitivity is 56% and final diagnosis can be established only by surgery,^[13] it is reported that 66% of cases in new series can be diagnosed by ultrasonography.^[14] Today, such increase in preoperative diagnosis rates depends on basically two reasons: Firstly, developments on the quality of sonographic imaging have helped to detect abnormal indications at earlier periods, and secondly, ART practices have not only increased heterotopic pregnancy prevalence but also have contributed to remind the possibility of heterotopic pregnancy during ultrasonographic examination.^[1]

If there is no risk factor in spontaneous heterotopic pregnancies such as infertility, pelvic inflammatory disease, ectopic pregnancy history, the possibility of heterotopic pregnancy is not reminded since early period of ectopic pregnancy is asymptomatic and normal intrauterine pregnancy is observed in these pregnancies, and thus the diagnosis is delayed. In the first examination of our case performed at an external center, final diagnosis was not reached for such reasons and the patient was referred to our clinic with the pre-diagnosis of molar pregnancy.

Treatment method is determined by the hemodynamic condition of the patient, localization of ectopic pregnancy, expectation of the family about intrauterine pregnancy and the experience of surgeon. In patients with disordered hemodynamia, emergency laparotomy may be suggested or laparoscopic surgery may be suggested in the presence of or experienced anesthesia and surgery team.^[15] If the hemodynamia of patient is stable, conservative methods can be used on appropriate indications as well as preferring laparoscopy. In cases

where intrauterine pregnancy is not desired, systemic methotrexate can be used successfully.^[16] For cases where intrauterine pregnancy is desired, it is reported that injecting local potassium chloride, hypertonic solution or low dose of methotrexate into ectopic pregnancy sac terminates ectopic pregnancy without damaging intrauterine pregnancy.^[17-21] Especially in non-tubal heterotopic implantations such as cervical, cornual or cesarean scar, surgical treatment is maternally associated with high bleeding and associated hysterectomy risks. In these cases, any surgical operation (whether maternal risks are observed or not during surgical operation) on uterus causes intrauterine pregnancy to result in miscarriage at a high possibility. Therefore, local treatments mentioned above are the most suitable approaches in especially non-tubal heterotopic implantations.^[1,16,19-21]

The most basic characteristics distinguishing heterotopic pregnancy management from ectopic pregnancy management are the cases where intrauterine pregnancy is desired,^[1] because the treatment method to be chosen will determine directly the prognosis of intrauterine pregnancy in such a case. When intrauterine component of heterotopic pregnancy is compared to normal intrauterine pregnancy, unfortunately 2-3 times more miscarriage rate is observed.^[22] On the other hand, Survival rates reported as 48-51% reported in 1950s associated with prognosis of intrauterine pregnancy are reported as 69% nowadays.^[14,23] Furthermore, in cases where intrauterine pregnancy continues, it is reported that the risk of observing bad gestational outcomes such as low birth weight and preterm labor display no significant difference than normal intrauterine pregnancies.^[22]

Conclusion

In all first trimester pregnancies, especially ART pregnancies, referring with the indications of asymptomatic or groin pain and peritoneal irritation symptoms, differential diagnosis of heterotopic pregnancy should certainly be reminded even though normal intrauterine pregnancy is followed-up. It should be known that normal TV USG indications will not rule out the diagnosis of heterotopic pregnancy in symptomatic patients although TV USG has a significant role in the diagnosis of such cases. Early diagnosis and proper treatment method has an essential significance in terms of maintaining intrauterine pregnancy healthily and protecting fertility of cases.

Conflicts of Interest: No conflicts declared.

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