Advanced Isthmic Ectopic Pregnancy Endangering the Life of the Mother: Case Report

Anne Yaşamını Tehdit Eden İleri İstmik Ektopik Gebelik

Ümit GÖRKEM,^a Musa ZORLU,^b Dilek YILMAZ,^c Cihan TOĞRUL,^a Tayfun GÜNGÖR^a

Department of Obstetrics and Gynecology, Department of General Surgery, Hitit University Faculty of Medicine, Clinic of Pathology, Hitit University Corum Training and Research Hospital, Corum

Geliş Tarihi/*Received:* 06.11.2015 Kabul Tarihi/*Accepted:* 20.06.2016

This Study was presented as a poster at 13. National Obstetricland Gynecology Congress, 11-15 May 2015, Antalya

Yazışma Adresi/Correspondence: Ümit GÖRKEM Hitit University Faculty of Medicine, Department of Obstetrics and Gynecology, Çorum, TURKEY/TÜRKİYE drumitgorkem@hotmail.com **ABSTRACT** Herein, we report a case of an advanced isthmic pregnancy endangering the life of the mother. A 23-year-old woman, gravida 2, para 1, was being routinely examined at the university polyclinic, when a precipitous deterioration of vital signs developed. An urgent laparotomy was undertaken, and a live isthmic pregnancy at 16 weeks of gestation, leading to massive intra-abdominal hemorrhage, was revealed. An accurate diagnosis had been missed at an early stage of the pregnancy. In contrast to the present case, isthmic ectopic pregnancies mostly rupture at early weeks. Continuation of pregnancy with a live fetus is a rare event in isthmic ectopic cases. Clinicians must be aware of the implantation site at an early stage of gestation to avoid a misdiagnosis as an intrauterine pregnancy at later stages.

Key Words: Pregnancy, ectopic; early diagnosis

ÖZET Biz bu yazıda anne yaşamını tehdit eden ileri istmik gebelik olgu sunumunu bildirmekteyiz. 23 yaşında, gravida 2 para 1, bir kadının üniversite polikliniğinde rutin muayenesi yapılmakta iken hastanın vital bulgularında ani bir bozulma gelişti. Acil laparotomi gerçekleştirildi ve yoğun intraabdominal kanamaya neden olan 16 gebelik haftasında bir canlı istmik gebelik saptandı. Doğru tanı gebeliğin erken dönemlerinde gözden kaçırılmıştı. Sunulan olgumuza zıt olarak istmik ektopik gebelikler çoğunlukla erken haftalarda rüptüre olurlar. İstmik ektopik olgularda canlı bir fetüs ile gebeliğin devamı nadir bir olaydır. Klinisyenler, ileri dönemde intrauterin gebeliğin yanlış tanı konulmasından kaçınmak için gebeliğin erken döneminde implantasyon yerinin farkında olmalıdırlar.

Anahtar Kelimeler: Gebelik, ektopik; erken tanı

Turkiye Klinikleri J Case Rep 2016;24(4):319-22

he incidence of ectopic pregnancy reaches nearly 1.5–2% of all pregnancies. Although the incidence has risen about six-fold over the past 30 years, the risk of death related to ectopic pregnancy has been reduced by almost 90%.¹ The highest mortality rate occurs at 15–19 years of age.² However, the likelihood of death due to an ectopic pregnancy is still higher than that from a live birth. An ectopic pregnancy mostly has a classical triad of symptoms: delayed menses, vaginal bleeding, and lower abdominal pain. The other symptoms associated with a ruptured ectopic pregnancy are shoulder pain, lightheadedness, and shock. Interference with normal tubal transport mechanisms may predispose to ectopic pregnancy. It is possible, but unproven, that endocrine factors predisposing to premature implantation take part in the pathogenesis of ectopic pregnancy.³ The

doi: 10.5336/caserep.2015-48542

Copyright © 2016 by Türkiye Klinikleri

Görkem ve ark. Kadın Hastalıkları ve Doğum

most common site of ectopic implantation is a fallopian tube, accounting for 98% of all ectopic pregnancies. Overall, 70% of ectopic pregnancies are implanted in the ampulla, 12% are implanted in the isthmus, 11% are implanted in the fimbria, and 2% are implanted in interstitial (cornual) segments. ^{4,5} Ovarian, abdominal, and cervical locations are relatively rare.

Herein, we present an unusual case of a live 16-week isthmic ectopic pregnancy endangering the life of the mother.

CASE REPORT

A 23-year-old woman, gravida 2, para 1, was admitted to the university polyclinic for a routine pregnancy examination. She had a gestation period of 16 weeks and 5 days according to the last menstrual period and was suffering from right-sided lumbar pain. Her medical history did not report any risk factors for ectopic gestation. Abdominal examination revealed a tenderness to palpation in the right upper and lower quadrants of the abdomen without guarding and rebound tenderness. Her blood pressure and heart rate were in the normal ranges, 100/75 mmHg and 92 bpm, respectively. Her complete blood count showed a normal hemoglobin level of 12.1 g/dL and normal levels of leukocytes and platelets. The ultrasonographic examination confirmed a live intrauterine pregnancy at 16 weeks and 3 days of gestation. On the ultrasonogram, a slight dilatation and a crystalloid appearance of the right maternal renal pelvis were detected. It was decided to refer the case for consultation to an urologist. Thereafter, her skin became pale, and the patient suffered from a shortness of breath in a short time. She was urgently transferred to the emergency department. The blood pressure reading was in the normal range, but the heart rate increased to 125 bpm. A second ultrasonography was performed, and an accumulation of fluid in the maternal abdominal cavity and a live intrauterine fetus were reported. A repeat test showed that hemoglobin dropped to 7.1 g/dL. The patient gave the written informed consent before surgical intervention. An urgent laparotomy was decided upon to determine the cause

of the clinical condition.

Under general anesthesia, a median laparotomy was performed, which revealed 2.5 L of intraabdominal blood. In addition, there was an unruptured right-sided ectopic pregnancy, which was located in the isthmic portion of the tube and bled from the fimbrial end (Figures 1, 2). A total right salpingectomy was performed. When the excised fallopian tube was opened, a male fetus weighing 96 grams, with normal external appearance, was delivered (Figure 3). Four units of erythrocyte suspension were administered to the patient. The postoperative period was uneventful, and the patient was discharged on the 10th day of hospitalization.

The histological results revealed that the development of the fetus was compatible with 16–17 weeks of gestation. The results of examination of fetal organs were reported to be normal. A microscopic study revealed chorionic villi and trophoblastic cells in the tubal lumen and attenuation of the muscular layer of the tuba uterine (Figure 4).

DISCUSSION

The present case is a rare example of isthmic ectopic pregnancy, which was viable at 17 weeks of gestation after a spontaneous conception. We were urged to perform a laparotomy to determine the reason why the life of the mother was at risk.



FIGURE 1: A view of unruptured right-sided ectopic pregnancy at the isthmic portion of tube.

Görkem et al. Gynecology and Obstetrics



FIGURE 2: Another view of unruptured right-sided ectopic pregnancy at the isthmic portion of tube.



FIGURE 3: The male fetus within the isthmic ectopic pregnancy.

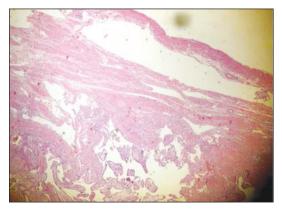


FIGURE 4: Histological findings of the excised specimen, the chorionic villi and trophoblastic cells in tubal lumen and attenuation of muscular layer of tuba uterina, (HE staining, original magnification X 40).

Few risk factors have been described that predispose women to ectopic pregnancy, but 50% of the women do not show them.^{6,7} Some of the reported risk factors are previous ectopic pregnancy, tubal surgery, intrauterine device, tubal pathology, infertility, the use of assisted reproductive technology, previous genital infections, smoking, and prior cesarean delivery. However, our case had not used any contraception method, and had no history of the other risk factors.

The histopathology of ectopic pregnancies shows a variety of implantation sites. Ectopic implantation in the tubal isthmus typically results in penetration of the tubal wall at a relatively early stage. In contrast, ampullary and especially interstitial ectopic implantations mostly lead to delayed penetration of the involved tubal segment, because the more muscular segments are less distensible.8 As a rule, a tubal rupture in the first weeks of pregnancy gives an impression that the ectopic pregnancy is situated in the isthmic portion of the tube. When a fertilized ovum is implanted within the interstitial portion, rupture occurs after 14-16 weeks and is usually spontaneous or occurs after coitus or a bimanual examination. There are few cases of advanced isthmic pregnancies reported in the literature. Noteworthy, the ectopic gestation in the present case progressed to 17 weeks without any clinical warning signs.

Ultrasonography is the usual diagnostic procedure of choice in most cases. The confirmation of an intrauterine pregnancy is performed by observing the uterine wall surrounding the fetus and placenta. Occasionally, sonographic findings may be vague, depending on examiner's experience and the quality of ultrasound equipment. Diagnosis of advanced ectopic gestations is only possible in 50-90% of unsuspected cases. However, even trans vaginal sonography cannot reveal ectopic implantation in up to 10% of the cases. In advanced ectopic cases, surgical intervention is essential for diagnosis and treatment to prevent likely dangerous hemorrhage. 9,10 In our case, an appropriate diagnosis of the implantation site was missing even at the late stage of gestation.

The incidence of uterine congenital anomalies due to Müllerian defects in normal fertile women

Görkem ve ark. Kadın Hastalıkları ve Doğum

is 3,2%. Unicornuate uterus is thought to be seen in 2,4-13% of all Müllerian anomalies.11 The pregnancy in a rudimentary horn (RH) is estimated between 1/76.000 and 1/140.000.12 In most cases, the gestational duration is longer than a tubal pregnancy. As the uterine wall in RH is thicker and has the variable musculature, the risk of uterine horn rupture is near to 50% especially during second and third trimester. Tubal pregnancy, cornual pregnancy, intrauterine pregnancy and abdominal pregnancy are common sonographic misdiagnosis. 13 Tsafrir et al. proposed the following criteria for ultrasonographic diagnosis: (1) a pseudo pattern of an assymetrical bicornuate uterus, (2) absent visual continuity tissue surrounding the gestational sac and the uterine cervix, and (3) the presence of myometrial tissue surrounding the gestational sac.14 In a review, sensitivity of sonography for diagnosis of RH pregnancy was estimated at 26%. RH pregnancy should be always be considered as a diffential diagnosis of tubal pregnancy. A tubal pregnancy will not have a ring of myometrium surrounding the gestational sac. Additionally, hypervascularity typical to placenta accreata may support the diagnosis of RH pregnancy.¹⁵

In conclusion, if an isthmic pregnancy is not diagnosed at an earlier gestational age, it poses a risk of potentially catastrophic intra-abdominal bleeding. Therefore, an urgent laparotomy becomes a necessity for hemodynamically unstable patients. ^{9,10} Clinicians must be aware of the implantation site at an early stage of gestation to avoid a misdiagnosis as an intrauterine pregnancy at later stages.

Acknowledgements

The authors would like to thank to Figen Yavuz, MD that helped to edit the manuscript.

REFERENCES

- Goldner TE, Lawson HW, Xia Z, Atrash HK. Surveillance for ectopic pregnancy--United States, 1970-1989. MMWR CDC Surveill Summ 1993;42(6):73-85.
- Tay JI, Moore J, Walker JJ. Ectopic pregnancy. BMJ 2000;320(7239):916-9.
- Attar E. Endocrinology of ectopic pregnancy.
 Obstet Gynecol Clin North Am 2004;31(4):779-94, x.
- Breen JL. A 21 year survey of 654 ectopic pregnancies. Am J Obstet Gynecol 1970;106(7):1004-19.
- Bouyer J, Coste J, Fernandez H, Pouly JL, Job-Spira N. Sites of octopic pregnancy: a 10 year population-based study of 1800 cases. Hum Reprod 2002;17(12):3224-30.
- Banhart KT, Sammel MD, Gracia CR, Chittams J, Hummel AC, Shaunik A. Risk factors for ectopic pregnancy in women with symptomatic first-trimester pregnancies. Fertil Steril

- 2006;86(1):36-43.
- American College of Obstetricians and Gynecologists. AGOC Practice Bulletin No: 94: Medical management of ectopic pregnancy. Obstet Gynecol 2008;111(6):1479-85.
- Senterman M, Jibodh R, Tulandi T. Histopathologic study of ampullary and isthmic tubal ectopic pregnancy. Am J Obstet Gynecol 1988;159(4):939-41.
- Cunningham FG, Leveno KJ, Bloom SL, Huth JC, Rouse DJ, Spong CY. Ectopic pregnancy. Williams Obstetrics. 23rded. NY: McGraw Hill; 2010. p.239-40.
- Kocak I, Dagdemir A, Yilmaz MS, Ustün C. Advanced tubal pregnancy at 30 weeks. J Obstet Gynaecol Res 2005;31(4):341-3.
- Simón C, Martinez L, Pardo F, Tortajada M, Pellicer A. Müllerian defects in women with normal reproductive outcome. Fertil Steril 1991;56(6):1192-3.

- Nahum GG. Rudimentary uterine horn pregnancy. A case report on surviving twins delivered eight days apart. J Reprod Med 1997;42(8):525-32.
- Bahadori F, Borna S, Behroozlak T, Hoseini S, Ayatollahi H. Failed induction in second trimester due to pregnancy in an uncommunicated rudimentary horn: case report. J Family Reprod Health 2009;3(3):95-7.
- Tsafrir A, Rojansky N, Sela HY, Gomori JM, Nadjari M. Rudimentary horn pregnancy: first trimester prerupture sonographic diagnosis and confirmation by magnetic resonance imaging. J Ultrasound Med 2005;24(2):219-23.
- Mohsin H, Khan MN, Jadun CK, Tanveer-ul-Haq. Role of ultrasound in detection of ectopic pregnancy: our experience. J Coll Physicians Surg Pak 2001;11:387-8.