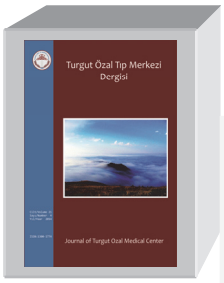

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Unembriyonic Pregnancy in a Noncommunicating Rudimentary Horn of a Unicornuate Uterus: A Case Report

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Abstract
This is a case report of an unembriyonic pregnancy in the non-communicating rudimentary horn of a unicornuate uterus with the contralateral ovary carrying a corpus luteum. A 24-year-old nullipara was admitted to our outpatient clinic with the complaint of primary infertility for 3 years. Her hysterosalpingographic and laparoscopic evaluation revealed a mullerian anomaly; non-communicating rudimentary uterine horn. One year later, the patient was admitted with a 10-week amenorrhea. The examination showed unembriyonic pregnancy in the left uterine rudimentary horn and corpus luteum on the contralateral ovary. The case at hand is an evidence of transperitoneal migration of both the sperm and the ovum, so the belief that unicornuate uterus with a non-communicating rudimentary horn goes asymptomatic without causing any trouble is not true. In such cases, the rudimentary horn should be removed.
Key Words: Mullerian Duct Anomaly; Pregnancy; Hysterosalpingography.

Unikornuat Uterusun Non-Communicating Rudimenter Hornunda Boş Gebelik: Vaka Takdimi

Özet
Çalışmamızda kontrolateral overde korpus luteum taşıyan, unikornuat uterusun non-communicating rudimenter hornunda oluşan unembriyonik gebelik sunulmuştur. Yirmi dört yaşında, daha önce gebelik geçirmemiş hasta üç yıllık primer infertilite şikayeti ile polikliniğimize başvurdu. Histerosalpingografik ve laparaskopik değerlendirilmesi müllerian bir anomaliyi; non-communicating rudimenter horn'u gösterdi. Bir yıl sonrası, hasta 10 haftalık adet gecikmesi ile başvurdu. Sol uterin rudimenter horn da unebmryonik gebelik ve karşı tarafta olan over de ise korpus luteum görüldü. Bu sunulan vaka, sperm ve ovumun her ikisinin de transperitoneal migrasyonun delilidir, non-communicating rudimenter horn'lu unikornuat uterusun herhangi bir sorun olmaksızın asemptomatik gideceği inanışı doğru değildir. Bu gibi vakalarda rudimenter horn çıkarılmalıdır.
Anahtar Kelimeler: Müllerian Kanal Anomalisi; Gebelik; Histerosalpingografi.

INTRODUCTION

The incidence rate of congenital uterine anomalies is difficult to determine since many women with such anomalies are not diagnosed, especially if they are asymptomatic. In a review of literature, the mean age of presentation is mid 20s (1). Uterine anomalies occur in 2% to 4% of fertile women with normal reproductive outcomes and unicornuate uterus is one of the least common congenital uterine anomalies (2-4).

There are three common developmental defects of the mullerian system: 1-Agenesis, 2-Lateral fusion defects, and 3-Vertical fusion defects.

The unicornuate uterus is an example of an asymmetric lateral fusion defect and occurs with an incidence of 1:4020 women in the general population (5). The cavity is usually normal, with a fallopian tube and cervix, while the failed mullerian duct has various configurations. The affected mullerian duct may not develop at all, or it may develop only partially either as a rudimentary horn on the uterus or as an anlage (a cluster of embryonic cells) (1). This horn or anlage may or may not communicate with the uterus. If the rudimentary horn is obstructed (without communication to the other uterus or cervix), patients may develop cyclic or chronic abdominopelvic pain and require surgical excision of the obstructed horn.

Ectopic pregnancy, first trimester abortion, second trimester abortion, preterm delivery, intrauterine fetal demise, and live birth rates in unicornuate uterus have been reported to be 2.7%, 24.3%, 9.7%, 20.1%, 10.5%, and 49.9%, respectively (5, 6).

Women with a unicornuate uterus are at higher risk for infertility, endometriosis, premature labor, and breech presentations (7).

Several cases of ruptured rudimentary horn pregnancies and ectopic tubal pregnancies in unicornuate uterus with rudimentary horn have been reported previously (8-10), but only one case of unembriyonic pregnancy in the non-communicating rudimentary horn with the contralateral ovary carrying a corpus luteum could be identified among all these reports (11). This paper reports such a rare case.

CASE REPORT

A 24-year-old nullipara was admitted to the outpatient clinic with primary infertility complaint for 3 years. She had menarche at 13. Her hysterosalpingographic evaluation revealed a mullerian anomaly; a single uterine cavity with only one tube, suggesting a unicornuate uterus (Figure 1B) and within the same month she underwent a laparoscopic evaluation. In the laparoscopy, the uterus was situated on the right. There were normal adnexa and a round ligament at the right side of the uterus, but none at the left. The uterus was considered to be a unicornuate uterus. On the left side, we observed a non-functional 2x3cm rudimentary uterine horn. This horn was connected by a fibrous strand to the internal os of the uterus and it contained the left round ligament (Figure 1A). The right fallopian tube and ovary were normal. The left ovary was adhered to the lateral abdominal wall and only the fimbrial portion of the left fallopian tube could be seen. Methylene blue passed only through the right tube whereas no fluid passage was observed through the left tube.

The patient underwent a laparoscopy one year later when she was re-admitted for 10-week long amenorrhea. Although β-hCG level was measured at 7557 IU/dL, the ultrasonography revealed no gestational sac in the endometrial cavity. An irregular shaped 30x24mm gestational sac without an embryo in the left rudimentary horn and a 30mm corpus luteum in the right ovary were also noted on the sonographic examination. A laparatomy was performed for the diagnosis of an unembriyonic pregnancy in the left uterine rudimentary horn. The rudimentary horn was excised and the salpingectomy of the left tube was performed. The pathologic examination revealed the presence of placental tissue and these findings were associated with the pregnancy (Figure 1C).

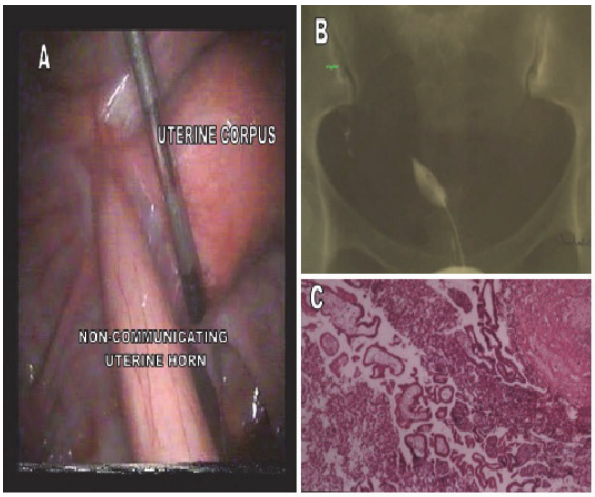

Figure 1. A. Rudimentary uterine horn with no communication. **B.** Hysterosalpingographic view. **C.** Choriovillic structures in the tissue circumscribed by the trophoblastic cells in the cross-section (hematoxylin and eosin, x 40).

DISCUSSION

The patient described in this report had an unembriyonic pregnancy in the non-communicating horn on the left along with both the corpus luteum and the unicornuate uterus with normal tube on the right. Previous reports confirmed the trans-peritoneal migration of either the oocyte or the sperm (11-13). In ectopic pregnancies, the presence of contralateral corpus luteum was reported in about 16% of cases (12). The ovum pickup was assumed to be accomplished by the opposite fallopian tube following trans-peritoneal migration of the oocyte in these cases. In patients with unicornuate uterus ectopic pregnancy in the contralateral non-communicating horn is also an evidence for trans-peritoneal sperm migration. In the literature, spontaneous ectopic pregnancy in a non-communicating heterotopic fallopian tube coexisting with corpus luteum in the contralateral ovary supports the hypothesis of transperitoneal migration of gametes or embryos (11,13).

There is no possibility that a normal pregnancy will occur via the non-communicating tube and that ectopic pregnancy is a definite risk in the heterotopic tube. Therefore salpingectomy is advised for the prophylaxis for ectopic pregnancies that sometimes result from trans-peritoneal migration of the sperm. However, according to the present literature, the effect of rudimentary horn on the gestational capacity in a patient with a unicornuate uterus is not clear. Furthermore, it is suggested that removal of the rudimentary horn eliminates the irritating action created over the main hemi-uterus during gestational expansion and the risk of peritoneal endometriosis (7).

Life threatening risks in cases of non-communating rudimentary horn pregnancies are described in the literature. The first case of uterine rupture associated with rudimentary horn was reported in 1669 by Mauriceau (14). The timing of rupture varies from 5 to 35 weeks depending on the horn musculature and its ability to hypertrophy and dilate. 70–90% ruptures before 20 weeks can be catastrophic (15). As the uterine wall is thicker and more vascular, bleeding is more severe in rudimentary horn pregnancy rupture (16). Kadan and Romano described rudimentary horn rupture as the most significant threat to pregnancy and a life-threatening situation (17). Maternal mortality rate before 1900 was reported to be 47.6%. Rupture of the horn is still common but no case of maternal death has been published since 1960 (18). Early diagnosis of the condition is essential and can be challenging. Ultrasound, hysterosalpingogram, hysteroscopy, laparoscopy, and MRI are diagnostic tools (19).

The present case disproof the belief that unicornuate uterus with a non-communicating rudimentary horn goes asymptomatic without causing any trouble. This case shows that, in addition to the previously mentioned risks of rupture of the horn, ectopic pregnancy, and the irritating action of the rudimentary horn over the hemi-uterus, rudimentary horn pregnancy in the contralateral

non-communicating horn is another problem and seen rarely. In the light of this data, it would be a prudent approach to remove the rudimentary horn when diagnosed incidentally during laparoscopy or laparotomy.

This study has been presented in Turkish-German Gyneocology Congress, 17 May 2005, Antalya.

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