

## A Rare Case of Bilateral Ectopic Pregnancy and Differential Diagnosis of Gestational Trophoblastic Disease

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### Abstract

**Background:** Bilateral ectopic pregnancy is a rare condition and is divided in two subgroups, primary and secondary, based on history of assisted reproductive technology.

**Case Presentation:** A 30 year old primigravid woman with history of infertility and ovulation induction presented to a hospital in Kashan in year 2013. She had vaginal bleeding, abdominal pain and ultrasound findings suggested early pregnancy. Due to high titer of  $\beta$ -HCG, gestational trophoblastic disease was proposed and D&C was done in referral and admission to gynecology ward in Tehran. Repeat sonography suggested ectopic pregnancy in left side and repeat  $\beta$ -HCG level showed an increase of 19435 *mIU/ml*. Laparotomy findings revealed bilateral ampullary ectopic pregnancy. Bilateral salpingostomy followed by one course of methotrexate was prescribed.

**Conclusion:** Bilateral ectopic gestation should be considered as a rare differential diagnosis for ectopic pregnancy. In this study, bigger size and rupture in left side was observed.

**Keywords:** Ectopic pregnancy, Reproductive techniques, Rupture.

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### Introduction

The implantation and development of fertilized ovum outside the uterine cavity is observed in approximately 2% of all pregnancies. Over 95% of ectopic pregnancies develop in fallopian tubes and usually in ampullary part. Because of the increase in incidence of sexually transmitted diseases, tubal surgery and more frequent use of ovulation induction and assisted reproductive technologies (ART), the incidence of ectopic pregnancies has grown in the last 30 years according to many reports from developed countries. Ectopic pregnancy is still the leading cause of pregnancy-related deaths in developed countries (1).

However, bilateral tubal pregnancy is a rare clinical condition which occurs in only 1 per 200,000

pregnancies. This condition is divided in two subgroups, primary and secondary, and because of different physiopathological mechanisms, these two entities should be studied separately. Incidence of each of these subgroups is about 50%. However, as studies have shown, the cause of bilateral ectopic pregnancy after ART is clearly different from spontaneous cases. It seems that previous tubal disease is a common risk factor in both situations. Clinical and paraclinical findings of this condition are the same as unilateral ectopic pregnancy so the distinct diagnosis is difficult (2).

In the present article, a case of bilateral tubal pregnancy is presented besides reviewing the relevant literature.

### Case Presentation

A 30 year old primigravid woman has been hospitalized in Kashan in year 2013 due to minimal vaginal bleeding and low abdominal pain. She had history of 2 years of primary infertility. She had been under different treatments including sequential treatment with clomiphene, FSH and HMG in previous cycle. Her last menstrual period was 8 weeks ago. In admission, vital signs were normal. The first transvaginal ultrasound revealed a suspicious gestational sac without yolk sac in uterus and normal adnexal area was reported. Due to high level of  $\beta$ -HCG (16000 *mIU/ml*), without normal viable intrauterine pregnancy, dilatation and curettage had been done to clarify existence of intrauterine pregnancy or its complications or EP. Due to high level of  $\beta$ -HCG, gestational trophoblastic neoplasm was considered in differential diagnosis besides ectopic pregnancy and it resulted in gynecology-oncology consultation. Pathologic report of endometrial sample revealed the existence of decidual tissue. The rise of  $\beta$ -HCG level to 21770 *mIU/ml*, 9 days after operation resulted in referral and admission of patients to gynecology-oncology ward of our center in Tehran.

Positive findings in this admission to gynecology-oncology ward in Tehran were low abdominal pain, minimal vaginal bleeding with stable vital signs similar to the ones observed in previous admission. Through re-checking the  $\beta$ -HCG level, titer of 19435 *mIU/ml* was observed. The next ultrasound reported a normal uterus with endometrial thickening up to 11 *mm*, a heterogeneous mass measuring 49 *mm* in left adnex, suspicious to ectopic pregnancy and two simple cysts, 69 *mm* in left ovary and 50 *mm* in right ovary. There was no free fluid in the pelvic cavity. Because of persistently high level of  $\beta$ -HCG, surgical intervention was done. Atypical findings and diagnosis of GTN besides FP led to choice of laparotomy instead of laparoscopy. Surgical findings were two bluish, 5 *cm* in left side and 2 *cm* in right side, swelling in both ampullary parts of fallopian tubes with minimal bleeding from fimbria. Two simple cysts, about 5 *cm* in right ovary and 4 *cm* in left ovary were seen. Aspiration was done before surgical incision and clear serous fluid was aspirated from cyst. Bilateral salpingostomy was performed and some trophoblastic-like tissue was extracted and sent for pathologic evaluation separately. The patient received 50 *mg/m<sup>2</sup>* intramuscular methotrexate due to bipateral FP and non-radical ap-

proach. Trophoblastic tissue was reported in pathologic exam of sample sent from tubal ampullar lumen.  $\beta$ -HCG titerage revealed a decreasing trend in subsequent examinations.

### Discussion

Secondary bilateral ectopic pregnancy is a condition with localization of trophoblastic tissue in both tubes, following a kind of manipulation in physiology such as through the use of ART drugs. The literature and clinical findings regarding diagnosis of bilateral ectopic pregnancy are not comprehensive. The classic triad of pain, vaginal bleeding and missed period was present, similar to unilateral ectopic pregnancy. In a study, 19 secondary bilateral tubal pregnancy cases were reviewed in an attempt to examine the symptoms and signs. Thirteen out of 19 (68.4%) revealed a silent clinical course. Diagnosis in this group was done due to discrepancy between sonography and  $\beta$ -HCG level. In 6 out of 19 (31.6%) cases, hypovolemic shock due to rupture of gestation was the presenting feature. Due to involvement of both fallopian tubes, probability of rupture is higher in comparison to unilateral ectopic gestation (2). In the case of the present study, as a secondary bilateral ectopic gestation, high level of  $\beta$ -HCG without intrauterine pregnancy suggested ectopic gestation and atypical high titer of HCG resulted in referral to a tertiary gynecology-oncology ward. In review of studies, mean gestational age at the time of diagnosis was about 6.7 weeks (5-9 weeks) after the last menstruation (2).

Sonographic findings and  $\beta$ -HCG level, usually, don't lead to a correct diagnosis of bilateral ectopic pregnancy. Due to ample amount of  $\beta$ -HCG during normal gestation, its level is not diagnostic (1-3). In a review of HCG level in 16 cases of secondary bilateral tubal pregnancy, mean level of  $\beta$ -HCG was 20878 *mIU/ml* with a wide range of 27-226768 *mIU/ml*. Three cases out of 16 revealed  $\beta$ -HCG titers of 13296, 62520 and 226768 *mIU/ml* (2, 4-7). In unilateral ectopic pregnancy, there is lower level of  $\beta$ -HCG. In contrast to normal pregnancy with predictable range of  $\beta$ -HCG in each week of gestation, in ectopic pregnancy it is more variable (2, 3). In our case, empty uterus coexisting with high level of  $\beta$ -HCG suggested gestational trophoblastic disease (GTN) and ectopic pregnancy was less probable due to high atypical titer 21770 *mIU/ml* HCG. High level of  $\beta$ -HCG titer in bilateral ectopic gestation, such as

**Table 1.** Size of right and left tube distention according to surgical findings in cases with bilateral ectopic pregnancy

Published cases	Side of rupture	Size of R.T	Size of L.T
Weder et al. (1956) (11)	Left side	Pea-size	--
	Left side	Marble-size	--
Geiger et al. (1971) (12)	Right & left side	--	4 cm
Brady et al. (2005) (13)	Left side	Small size	
Ghaffari et al. (2011) (4)	--	small	3 cm
Sreeja Rani V.R et al. (10)	--	3*3cm	5*4 cm

above 10,000 in 3 case reports and our case and even 226768 *mIU/ml* in a case report, proposed GTN as a reasonable differential diagnosis. However, pre-operative diagnosis of secondary bilateral ectopic pregnancy was made just in 10% of patients (2). In our case, diagnosis was made during the operation, too. Pre-operative impression was adnexal mass probably coexisting with unilateral ectopic pregnancy.

Based on intra-operative findings, most of patients had ampullary ectopic pregnancy. Bilateral rupture of tubes was rare. In 5 out of 19 cases in a review of secondary bilateral ectopic pregnancy, unilateral tubal rupture in operative field was demonstrated. None of cases were bilaterally ruptured (2). In our case, bilateral unruptured tubal pregnancy in ampullary portion of tubes was seen. In our case, left tube was more distended, about 5 cm and right tube was 2 cm. In the review of bilateral tubal gestation in literature including primary and secondary cases, left tubal pregnancy was larger and more prone to rupture in comparison to right one. An attempt was made to review cases of medical literature which the side and size of rupture in bilateral tubal gestation was defined (Table 1). Medical treatment of these patients is not fully studied when pre-operative diagnosis is made. Medical management should involve the use of methotrexate. There are three surgical approaches for ectopic pregnancy: salpingotomy, salpingostomy and radical salpingectomy. Regarding the future plan of fertility procedures, conservation or resection of fallopian tubes might be planned in the operation field (1, 2, 8-10). In this case, bilateral salpingostomy was done.

### Conclusion

Bilateral ectopic gestation, although rare, should be regarded as the differential diagnosis for ectopic pregnancy, especially in assisted reproductive

technology cases and high titers of  $\beta$ -HCG (more than 100000). Left tubal side of bilateral tubal pregnancy was more probable to rupture and it revealed bigger size in review of cases.

### Conflict of Interest

The authors declare no conflict of interest.

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