DOI: https://doi.org/10.2298/SARH221227086B
UDC: 618.31

CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Spontaneous bilateral tubal pregnancy – case report and review of diagnostic and treatment difficulties

Jovan Bila[1,2], Jelena Dotlić[1,2], Ljubiša Jovanović[3], Milan Dokić[1], Tatjana Madić[1]

[1]University Clinical Center of Serbia, Clinic of Obstetrics and Gynecology, Belgrade, Serbia;
[2]University of Belgrade, Faculty of Medicine, Belgrade, Serbia;
[3]University Clinical Center of Serbia, Clinic of Obstetrics and Gynecology, Department of Pathology and Medical Cytology, Belgrade, Serbia

SUMMARY
Introduction Spontaneous bilateral ectopic pregnancy is a rare condition easily overlooked or misdiagnosed. We present a case of spontaneous bilateral tubal ectopic pregnancy and discuss the difficulties in diagnosing and treating such patients.
Case outline A 39-years-old patient with a history of irregular and abundant menstrual cycles complained of pelvic pain and light bleeding after 55 days of amenorrhea. Ultrasound revealed enlarged uterus with a myoma and a heterogenic formation with echogenic ring sign beside the left ovary. Free fluid with clots was present in the pelvis. As the patient was hemodynamically unstable and ectopic pregnancy was suspected, emergency laparotomy was performed. During the surgery we found that both tubes were significantly edematous, dilated, and livid in their ampullary regions. On the left tube anterior wall rupture 15 × 8 mm was noticeable, while right tube was intact with bleeding from its abdominal ostium. Even though the patient was not informed about the possibility of bilateral salpingectomy, after thorough consideration and due to the extent of tubal damage bilateral salpingectomy was eventually performed. Histopathological analysis confirmed the presence of decidua, partially viable and partially necrotic chorionic villi, and trophoblastic tissue in both right and left tubes.
Conclusions Careful preoperative and intraoperative examination of both Fallopian tubes as well as the whole abdominal and pelvic cavity should be mandatory during every assessment of patients with ectopic pregnancy.
Keywords: ectopic pregnancy; bilateral; spontaneous; diagnosis; treatment

INTRODUCTION

Ectopic pregnancy occurs in 1.5–2% of all pregnancies. Even with modern diagnostic and therapeutic tools ectopic pregnancy remains the most common life-threatening emergency in early pregnancy, causing significant maternal morbidity and mortality [1, 2, 3].

The ectopic trophoblast implantation mostly occurs in the ampullar region of one of the Fallopian tubes. However, two or more simultaneous gestations can occur in both of the tubes in the same patient [1, 2]. This condition is called bilateral ectopic pregnancy and is a rare clinical entity (1 in 200,000 pregnancies) usually associated with assisted reproduction. Contrary, primary i.e., spontaneous bilateral ectopic pregnancy is even more infrequent (1/725-1580 of the ectopic pregnancies) [1–4].

We present an incidental finding of spontaneous bilateral tubal ectopic pregnancy during urgent laparotomy and discuss the difficulties in diagnosing and treatment of such patients.

CASE REPORT

A 39-years-old patient was admitted to our hospital due to diffuse moderate blunt intermittent pelvic pain during the previous three days. Moreover, she had just started bleeding after the period of amenorrhea, what was accompanied by general weakness and coldish sweating.

The patient complained that her menstrual cycles were irregular (25–45 days), profuse and lasting 10–15 days since her last delivery. At the time of examination, amenorrhea lasted for 55 days. Still, patient stated that during this period of amenorrhea she noticed occasional spotting. There was no past history of any risk factor that might be linked to ectopic pregnancy (sexually transmitted diseases, pelvic inflammatory diseases, pregnancy termination, previous operations, intrauterine devices, fertility enhancing drugs or assisted reproduction). She had two spontaneous pregnancies followed by term vaginal deliveries of healthy children five and two years prior.

On examination she was pale, sweaty, and hypotensive with a blood pressure of 90/60 mmHg and a pulse of 92 beats per minute. The abdomen showed signs of peritoneal irritation (tension and painfulness). Her vulva was blood-stained and light fresh bleeding from the uterus was visible. Gynecologic examination revealed enlarged, softened uterus with uneven surface and bilateral thickening of adnexal regions. The uterus, adnexa, and the pouch of Douglas were

Received • Примљено: December 27, 2022
Revised • Ревизија: September 13, 2023
Accepted • Прихваћено: September 21, 2023
Online first: September 29, 2023

Correspondence to:
Jovan BILA
University of Belgrade
Faculty of Medicine
University Clinical Centre of Serbia
Clinic of Gynecology and Obstetrics
Dr Koste Todorovica 26
11000 Belgrade
Serbia
bilamsj@gmail.com

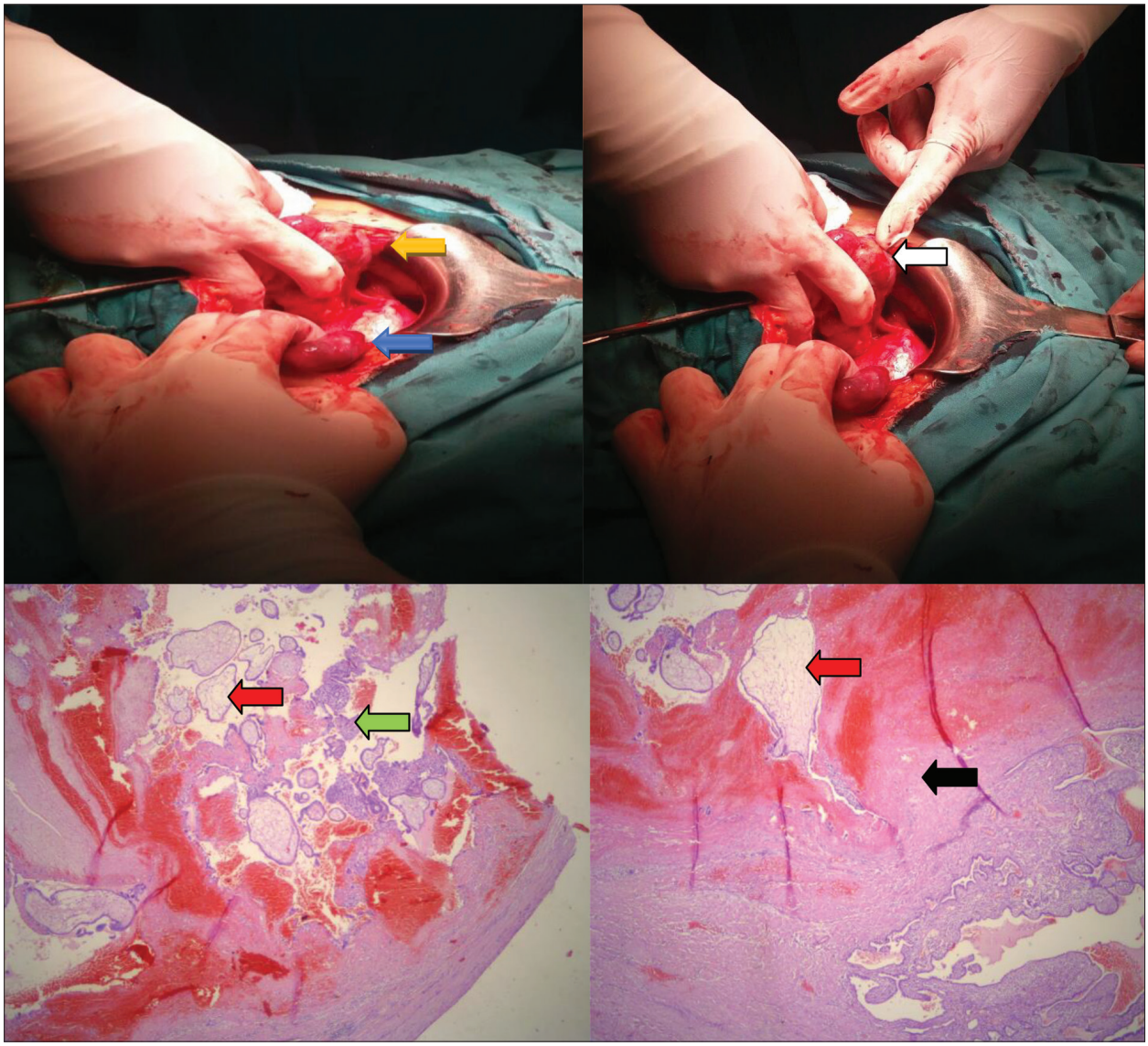

Figure 1. Intraoperative and histopathological findings of bilateral ectopic pregnancy: yellow arrow points to the left Fallopian tube with tubal pregnancy; blue arrow points to the right Fallopian tube with tubal pregnancy; white arrow points to the rupture of the anterior wall of the left Fallopian tube; red arrows point to extrauterine chorionic villi in both tubes; green arrow points to trophoblast; black arrow points to ectopic decidua attached to tubal wall

painful on palpation. There were no other pathological findings. Hemoglobin and hematocrit levels were significantly decreased (92 g/l and 30% respectively), but all other laboratory tests were in referral range. Serum Beta human chorionic gonadotropin (HCG) was 14,750 IU/l.

Ultrasound scan showed enlarged uterus 98 × 65 × 80 mm, with a myoma 43 × 35 mm on the right lateral and posterior wall. The endometrium was 14 mm thick with no signs of intrauterine gravidity. The left ovary was 36 × 23 mm in diameter with a follicle sized 19 mm. Next to the left ovary a heterogenic formation sized 39 × 35 mm with echogenic ring sign, suspected of ectopic gestational sack was seen. The right ovary was 34 × 32 mm in diameter with the largest follicle of 13 mm. Free fluid with blood cloths was presented in the Douglas pouch.

Considering the patient's condition, clinical, and ultrasound signs of ectopic pregnancy accompanied by abdominal hemorrhage, we performed an emergency laparotomy. The patient was informed about her condition and findings, the need for urgent surgery and the possibility of removing the left Fallopian tube. During surgery, we astonishingly found that both tubes were significantly edematous, dilated (right 37 × 32 mm and left 35 × 44 mm) and livid in their ampullar segments (Figure 1). Both tubes fell into the Douglas pouch and blood was dripping from the ruptured anterior wall (15 × 8 mm in diameter) of the left Fallopian tube. The right tube was intact but still small amount of blood was observed on the fimbrial end.

Because of the extent of tubal damage left salpingectomy had to be performed using Covidien LigaSure 5 mm Sealer/Divider® (Medtronic, Dublin, Ireland) coagulation. Right salpingotomy just above the dilated part of the tube and evacuation of ovular tissue from the Fallopian tube was attempted, but it was followed by abundant bleeding from the site of the salpingotomy. Consequently, after considering patients' hemodynamic instability and that she already

had two normal childbirths, it was decided to perform the right salpingectomy as well. Upon this, hemodynamic stability was achieved.

Histopathological analysis confirmed the presence of decidua, partially viable and partially necrotic chorionic villi, and trophoblastic tissue in both right and left tubes (Figure 1). The postoperative course was uneventful, and the patient fully recovered. The patient was informed about an unexpected finding and the course of the operation as this life-threatening condition required to extend the procedure, with which she consented.

The authors declare that the article was written according to ethical standards of the Serbian Archives of Medicine as well as ethical standards of medical facilities for each author involved. No personal data of the patient were presented in the manuscript.

The patient's written consent was obtained for the writing of this case report.

DISCUSSION

The typical risk factors for ectopic pregnancy include sexually transmitted diseases, endometriosis, and surgical intervention in the pelvis and abdomen that all can cause damage of the endosalpinx or adhesions that incorporate the Fallopian tubes. Also, Müllerian malformations, hormonal imbalances, late and multiple ovulations, endometrial tissue movements, tubal transmigration of the fertilized ovum, history of previous ectopic pregnancy, tobacco smoking, etc. [5, 6, 7]. In recent decades, one of the most common causes of ectopic pregnancy is assisted reproduction techniques especially those with ovulation induction. On the other hand, in the case of spontaneous bilateral tubal pregnancy, a spontaneous ovulation of two or more oocytes or the early embryo division must occur which is quite rare [4, 6]. Therefore, a personal or family history of twinning and the use of fertility drugs are considered to increase the risk of spontaneous ectopic pregnancy. Still, approximately half of women with ectopic pregnancies do not have any known risk factors [1, 2]. Our patient also had no major risk factors. Her only complaints were irregular cycles in past two years that implied hormonal disturbance. Her endocrinological status was still under the evaluation at the time of bilateral ectopic pregnancy occurrence. Moreover, abundant bleeding and anemia could imply on the cycle disturbances due to uterine myoma.

The majority of all ectopic pregnancies (up to 97%) are localized in the Fallopian tubes especially in their ampullar region (80%) [5]. The same is for bilateral tubal ectopic pregnancy that is ampullar in 73% of cases, and around 70% are unruptured at the time of diagnosis [1, 2, 8]. The gestational age of bilateral ectopic pregnancy at the time of diagnosis is usually 5–13 gestational weeks [1, 2, 8]. Our patient was in the sixth gestational week and had a typical, ampullar localization of ectopic pregnancy in both Fallopian tubes. Moreover, while one tube was intact, the other had already ruptured causing hematoperitoneum.

The most common symptoms/signs of ectopic pregnancy incorporate the triad of amenorrhea, vaginal bleeding, and pelvic pain. As the condition progresses with the rupture of the tubes, patients develop abdominal bleeding, acute abdomen, and finally hemorrhagic shock [6, 9]. The majority of patients with bilateral ectopic pregnancy have similar presentation and findings to those with a unilateral ectopic pregnancy, which makes the early diagnosing of bilateral tubal pregnancy particularly challenging [10–13]. Clinical analysis of our patient confirmed classical signs and symptoms of ectopic pregnancy. However, at that moment there were no indications for bilateral tubal affection. Moreover, due to irregular menstrual cycles the patient did not suspect pregnancy at all, which delayed the diagnosis.

Thorough gynecological examination with detailed transvaginal ultrasound scan can usually detect ectopic pregnancy in around 90% of cases [1, 2]. Color Doppler increases the ultrasound diagnostic accuracy even more [7, 8]. However, bilateral ectopic pregnancy is easily overlooked even when unilateral one is diagnosed due to the fact that ultrasound finding explains all of the clinical symptoms [4, 7]. Furthermore, different uterine and adnexal pathologies could mislead the interpretation of the ultrasonographic findings [12].

Moreover, the Beta HCG serum levels are not reliable in these cases. Generally, in case of ectopic pregnancy Beta HCG levels are increased, but their amplification progresses slower than in the healthy intrauterine pregnancy. Studies have also shown that in patients with bilateral ectopic pregnancy Beta HCG levels could be either significantly higher, or in the referral range for the gestational week, just like in our case [7, 8, 9].

Consequently, bilateral tubal ectopic pregnancy is usually confirmed only intraoperatively upon careful inspection of both tubes. Missed diagnosis could delay the treatment and worsen prognosis, making it a life-threatening condition [6, 7, 14]. In this case, we missed the ectopic pregnancy in the right tube by the transvaginal ultrasound as it was superposed by the myoma of the right uterine wall.

The treatment of ectopic pregnancy includes medications or surgical approaches that are selected according to patients' general condition, extent of tubal damage, and need for fertility preservation [6, 14, 15]. However, due to its rarity, currently there are no adequate protocols for bilateral tubal ectopic pregnancy management [6]. Although there were patients efficiently treated with Methotrexate, the success rate was not satisfactory because the levels of Beta HCG were higher than the permissible for Methotrexate application at the time of diagnosis [1, 5, 6]. It is still unclear whether the usual dose of Methotrexate used for unilateral tubal ectopic pregnancy should or should not be increased in the case of bilateral one [1, 5, 6].

According to current guidelines, surgical treatment of ectopic pregnancy should be performed in case of significant abdominal pain, adnexal mass of 35 mm or larger, positive fetal heartbeat, and/or Beta HCG levels over 5000 mIU/ml [4]. It includes salpingectomy (removal of the affected tube if it is ruptured and significantly damaged) or salpingotomy (conservative method of pregnancy removal with

tubal preservation when the tubes have remained intact and in patients who wish to preserve fertility). Laparoscopy is method of choice for majority of these patients [1, 4, 5, 6].

The choice of treatment for patients with bilateral tubal ectopic pregnancy in which both tubes are damaged is never easy, especially in nulliparous young women when diagnosis is intraoperatively established [11, 12, 13]. Thus, it is advisable that in all cases of ectopic pregnancies patients should be informed of all potential treatment options and complications, and their consent is obtained before surgery [1]. In patients with bilateral tubal ectopic pregnancy bilateral salpingostomy is performed in 42%, bilateral salpingectomy in 33%, and combination of salpingostomy and salpingectomy in 25% of cases, mostly by laparotomy in more urgent and complex situations [6, 14, 15]. We had to perform bilateral salpingectomy as the patient was hemodynamically unstable during the operation, and taking into account that she had already had two healthy children.

In conclusion, the presented case highlights the fact that careful preoperative and intraoperative examination of genital organs, both Fallopian tubes, as well as the whole abdominal and pelvic cavity should be routine and mandatory during assessment of all patients with ectopic pregnancy.

Conflict of interest: None declared.

REFERENCES

1. Damiani GR, Arezzo F, Muzzupapa G, Dinaro E, Cicinelli E. Extra-uterine bilateral tubal ectopic pregnancy. Updates Surg. 2020;72(4):1289–90. [DOI: 10.1007/s13304-020-00778-7] [PMID: 32347448]
2. Gathura JE, Elfeky A, McLaren R Jr, Herzog D, Grazi R. Spontaneous Bilateral Tubal Ectopic Pregnancy in a Low-Risk Patient: A Case Report with Implications for Preoperative Patient Counseling. Case Rep Obstet Gynecol. 2021;2021:5588869. [DOI: 10.1155/2021/5588869] [PMID: 34249378]
3. Nyakura M, Mhlanga FG, Madziyire M, Matshalaga S. Spontaneous bilateral tubal ectopic pregnancy: a case report. Pan Afr Med J. 2021;38:395. [DOI: 10.11604/pamj.2021.38.395.28771] [PMID: 34381539]
4. Iwe ABC, Nwafor JI, Asiegbu OG, Adebayo JA, Uche-Nwidagu BN, Ali VC. Spontaneous Ruptured Bilateral Tubal Ectopic Pregnancy Following Natural Conception: A Rare Case Report. J Hum Reprod Sci. 2021;14(2):196–9. [DOI: 10.4103/jhrs.JHRS_87_20] [PMID: 34316237]
5. Demircioglu F, Steiner PH, Rosendal BM, Winther LP, Khalil MR. Bilateral tubal pregnancy in a woman without risk factors. Ugeskr Laeger. 2023;185(29):V04230229. [PMID: 37539800]
6. Masten M, Alston M. Spontaneous Bilateral Ectopic Pregnancy Treated With Combination of Methotrexate, Unilateral Salpingectomy, and Unilateral Expulsion of Pregnancy. Cureus. 2022;14(9):e29031. [DOI: 10.7759/cureus.29031] [PMID: 36237762]
7. Eghbali E, Azari M, Jafarizadeh A, Alihosseini S. Spontaneous bilateral tubal ectopic pregnancy preoperatively diagnosed by the ultrasound: a case report. BMC Pregnancy Childbirth. 2023;23(1):125. [DOI: 10.1186/s12884-023-05458-z] [PMID: 36823553]
8. Farshidpour LS, Vinson DR, Durant EJ. Bilateral Tubal Pregnancies Presenting 11 Days Apart: A Case Report. Clin Pract Cases Emerg Med. 2023;7(1):11–5. [DOI: 10.5811/cpcem.2022.10.56910] [PMID: 36859329]
9. Rowlatt B, Saeed M, Chester J. A rare and unusual case of bilateral tubal ectopic pregnancies. J Obstet Gynaecol. 2022;42(4):713–4. [DOI: 10.1080/01443615.2021.1959531] [PMID: 34698604]
10. Kyejo W, Rubagumya D, Fidaali Z, Jusabani A, Kaguta M, Jaiswal S. Bilateral tubal ectopic gestation: Complication in a patient with previous ectopic pregnancy, rare case report. Int J Surg Case Rep. 2022;97:107470. [DOI: 10.1016/j.ijscr.2022.107470] [PMID: 35926381]
11. Yang Z, Shangguan L, Chen Y, Jiang R. Bilateral tubal pregnancy caused by taking emergency contraceptive pill in Chinese woman: A case report. Asian J Surg. 2023:S1015-9584(23)00978-8. [DOI: 10.1016/j.asjsur.2023.06.121] [PMID: 37429795]
12. Zeng X, Luo L, Ou-Yang YW. A spontaneous bilateral fallopian tube pregnancy with didelphic uterus: A case report. Medicine (Baltimore). 2021;100(2):e24291. [DOI: 10.1097/MD.0000000000024291] [PMID: 33466216]
13. Al Dus G, Alhamoud AU, Ata Allah N, Alabdalla J. Two embryos did not implant into the womb. A rare case of non-iatrogenic bilateral ectopic pregnancy (two-tailed tubal ectopic pregnancy) case report. Ann Med Surg (Lond). 2021;71:102840. [DOI: 10.1016/j.amsu.2021.102840] [PMID: 34745598]
14. Niviti S, Gokani KH. A Rare Case of Spontaneous Bilateral Ruptured Tubal Ectopic Pregnancy. J Obstet Gynaecol India. 2019;69(5):470–2. [DOI: 10.1007/s13224-019-01206-6] [PMID: 31598053]
15. Wang ZZ, Xu LW, Zhao L, Chen YJ, Liu HC, Yu EK. Simultaneous bilateral fallopian tubal pregnancy of a laparoscopic definite spontaneous unilateral ovulation: a case report. BMC Pregnancy Childbirth. 2022;22(1):110. [DOI: 10.1186/s12884-022-04453-0] [PMID: 35144595]

Спонтана билатерална тубарна трудноћа – приказ случаја и тешкоће у дијагностици и лечењу

Јован Била[1,2], Јелена Дотлић[1,2], Љубиша Јовановић[3], Милан Докић[1], Татјана Мадић[1]

[1]Универзитетски клинички центар Србије, Клиника за гинекологију и акушерство, Београд, Србија;
[2]Универзитет у Београду, Медицински факултет, Београд, Србија;
[3]Универзитетски клинички центар Србије, Клиника за гинекологију и акушерство, Служба за патологију и медицинску цитологију, Београд, Србија

САЖЕТАК

Увод Спонтана билатерална ектопична трудноћа је ретко стање које се лако превиди или погрешно дијагностикује. Представљамо случај спонтане билатералне тубарне ектопичне трудноће и разматрамо потешкоће у дијагностици и лечењу таквих пацијенткиња.

Приказ случаја Пацијенткиња стара 39 година са историјом неправилних и обилних менструалних циклуса жалила се на бол у карлици и оскудно вагинално крварење након 55 дана аменореје. Ултразвуком је утврђена увећана материца са миомом и хетерогена формација са ехогеним прстенастим одјеком поред левог јајника. У карлици је била присутна слободна течност са коагулумима. Пошто је пацијенткиња била хемодинамски нестабилна и сумњало се на ектопичну трудноћу, урађена је хитна лапаротомија. Током операције уочено је да су оба јајовода била едематозна, проширена и ливидна у својим ампуларним сегментима. На левом јајоводу је примећена руптура предњег зида 15 × 8 *mm*, док је десни јајовод био интактан, са крварењем из абдоминалног отвора. Иако пацијенткиња није била обавештена о могућности билатералне салпингектомије, због степена оштећења јајовода, после детаљног разматрања урађена је билатерална салпингектомија. Хистопатолошком анализом потврђено је присуство децидуе, делимично вијабилних и делимично некротичних хорионских ресица, као и трофобластног ткива у левом и десном јајоводу.

Закључак Код пацијенткиња са ектопичном трудноћом неопходна је пажљива преоперативна и интраоперативна евалуација како унутрашњих гениталних органа, тако и целе мале карлице и трбушне дупље.

Кључне речи: ектопична трудноћа; билатерална; спонтана; дијагноза; лечење