

Spontaneous Bilateral Tubal Ectopic Pregnancy: Case Report and Literature Review

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Abstract

Bilateral tubal ectopic pregnancy is the rarest form of ectopic pregnancy and happens once in every 200,000 pregnancies. Of twin pregnancies, it is the most uncommon. The incidence increases in women undergoing assisted reproduction techniques or ovulation induction. Clinical presentation is unpredictable and there are no typical clinical features to distinguish it from a unilateral ectopic pregnancy. Pre-operative diagnosis is difficult, and it's usually made surgically. Treatment options vary according to the location of the ectopic pregnancy, to the tubal damage extension and the necessity of fertility preservation. This work's objective is to report a bilateral spontaneous tubal ectopic pregnancy that happened in the metropolitan area of Belo Horizonte, Minas Gerais, in 2019 and to review the literature regarding the subject.

Publication History:

Received: October 29, 2022

Accepted: January 26, 2023

Published: January 28, 2023

Keywords:

Ovarian carcinoma, Low-grade serous ovarian carcinoma, Gynecological cancer

Introduction

The ectopic pregnancy, which happens when the blastocyst implants outside of the uterine cavity, has an incidence of 2% of all pregnancies and in 98% of the cases it is located in the fallopian tube [1]. It is the main cause of mortality during the first trimester of pregnancy [2] and represents 10% of all deaths related to complications of pregnancies in developed countries [2].

It's prevalence has been increasing in the last 30 years and many risk factors have been identified, such as the increase in sexually transmitted infections which alter the pelvic cavity, the increase in tubal interventions, the use of contraceptives, specially intrauterine devices, smoking, and pelvic surgeries [3].

The diagnosis is based on clinical findings, β -hCG serum levels and transvaginal ultrasound which will show fetal content outside of the uterine cavity. Treatment depends on maternal hemodynamic stability, gestational sac size and hormonal dosages, and can be expectant, with methotrexate, or surgical by laparoscopy or laparotomy in which a salpingostomy or a salpingectomy can be performed [4].

Bilateral ectopic pregnancy is a rare form of twin pregnancy that happens once every 200,000 pregnancies. It is the most uncommon of twin pregnancies, with heterotopic or single tubal twin pregnancy being more frequent. It can spontaneously occur bilaterally (43% of the reported cases since 1980) or it can be related to assisted reproductive technologies, hormonal modulation and tubal manipulation [5]. Bilateral ovulation is necessary for it to happen spontaneously, which is rare in human beings [5]. The first bilateral tubal ectopic pregnancy reported in Latin America happened in Mexico in 1993 [6].

The diagnostic criteria determined by Norris include chorionic tissue found in both fallopian tubes simultaneously [7]. Clinically, bilateral ectopic pregnancy cannot be differentiated from unilateral ectopic pregnancy, although there is greater risk of tubal rupture and hemorrhagic shock since both tubes are compromised and preoperative diagnosis is difficult [5].

The present work's objective is to report a case of bilateral spontaneous tubal ectopic pregnancy that happened in the metropolitan area of Belo Horizonte, Minas Gerais in 2019.

The report was conceded by the patient after signing an informed consent form granting her anonymity.

Case Report

Patient, 31 years old, history of 2 previous pregnancies, 1 cesarean delivery and 1 abortion, currently spontaneously pregnant with an estimated gestational age of 6 weeks and 4 days, as calculated by last menstrual period (April, 7th, 2019), admitted to the hospital in May 23rd, 2019, with complaints of intense pain in the hypogastrium and right lower quadrant, no bleeding, fever or urinary symptoms and no history of sexually transmitted infections. Upon physical examination of the abdomen, uterus was not palpable and painful decompression of the right lower quadrant was observed. The pelvic examination showed a closed uterine cervix, not painful at mobilization. Initial blood workup: β -hCG was 4800 mUI/mL; reactive C-Protein was 3,9 units; Hemoglobin was 11,4 units; urinalysis and urine Gram staining were unremarkable. Transvaginal ultrasound showed a para-ovarian mass in the right adnexal region with an estimated volume of 8,8 cm³ and no free fluid on the pelvic cavity. She was submitted to exploratory laparotomy in May 25th, 2019, in which blood was found upon opening of the abdomen and adnexal masses were identified in both fallopian tubes upon exploration of the cavity, ruptured on the left and whole on the right. It was performed bilateral salpingectomy and the material was sent to anatomopathological analysis. At microscopy, parietal and intraluminal hemorrhage were found in both tubes, as well as chorionic villi intermingled, and as a conclusion, bilateral ectopic pregnancy.

Discussion

Spontaneous bilateral ectopic pregnancy, or primary bilateral tubal ectopic pregnancy, is an extremely rare event, with an estimated

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Citation: Valadão CED, de Melo CSB, Brandão LV, Tofani GB (2023) Spontaneous Bilateral Tubal Ectopic Pregnancy: Case Report and Literature Review. Int J Gynecol Clin Pract 10: 165. <https://doi.org/10.15344/2394-4986/2023/165>

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incidence of 1/725 to 1/1580 cases of ectopic pregnancies, which is equivalent to 1 case for every 200,000 pregnancies [8].

The general number of bilateral ectopic pregnancies has been increasing as a result of the broadening and popularization of assisted reproductive technologies. Other risk factors include history of twin pregnancy in the family, sexually transmitted infections in the last decade, pelvic anatomy distortion due to endometriosis or surgical procedures, smoking, ovulation induction and progesterone only contraceptives which alter tubal motility [9].

In the reported case, the patient didn't present any of the risk factors, corroborating with a few studies in which half of women with ectopic pregnancies did not present any previous risk factor [10].

Numerous theories have been proposed to explain this phenomenon of simultaneous ectopic pregnancies. Among the most accepted ones are the double ovulation with implantation of the egg in areas of previous tubal lesion (superfertilization), the theory of transperitoneal trophoblastic migration of an extra-uterine pregnancy to the other fallopian tube, and lastly the fecundation of a second oocyte in an already pregnant woman (superfetation), it being difficult to define which phenomenon happens in each case [11].

Unlike unilateral ectopic pregnancy, non-invasive diagnostic methods such as seriated β -hCG dosage and sonography were shown to be faulty in diagnosing bilateral ectopic pregnancy. By revising 42 reported cases in the literature, it was realized that bilateral ectopic pregnancy is not correlated to serum β -hCG levels; in nearly all cases, hormone levels were correspondent to a single ectopic pregnancy or were even lower [5]. There are only two cases that report the diagnosis being made ultrasonographically. What usually happens is that when the examiner sees an ectopic pregnancy in one of the tubes, his/her attention is drawn to that pathology and other pertinent alterations may pass unseen, like a bilateral ectopic pregnancy. It is suggested then that all patients diagnosed with unilateral tubal ectopic pregnancy and who have risk factors should be advised before surgery on the possibility of bilateral tubal ectopic pregnancy and that therapeutic options should be discussed if this occurs [12].

Most bilateral ectopic pregnancies are then diagnosed during the intraoperative period and salpingectomy is usually the procedure performed [8]. Currently, laparoscopy is the gold standard for this diagnosis.

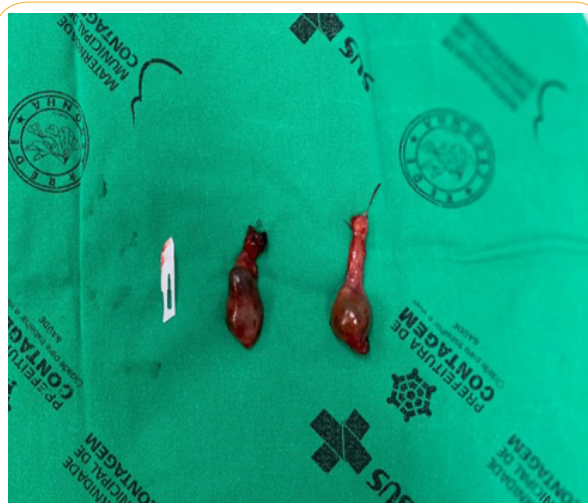


Figure 1: Shows bilateral ectopic pregnancy.

The case reported above reiterates the difficulty in diagnosing this pathology. Her serum β -hCG levels were compatible with a single pregnancy and it was not possible to demonstrate bilateral adnexal masses during transvaginal ultrasound. Thus, it is essential and mandatory the careful evaluation of the abdominal cavity and of both fallopian tubes as a routine during the surgical procedure in all cases of ectopic pregnancies because even without risk factors, the occurrence of bilateral tubal ectopic pregnancies simultaneously is possible, which, in most cases, are suspected and diagnosed only during surgery. There are cases reported in the literature in which ectopic pregnancy contralateral to the one identified on ultrasound passed unseen during laparotomy and the patient had to be reopened a few days later due to rupture [8].

Due to its rarity, there are no guidelines or specific protocols to the management of bilateral ectopic pregnancies. Usually, conduct varies according to patient's stability, tubal damage extension and the desire of a future pregnancy. Conservative management with methotrexate can be adopted, but to present date, there are no publications regarding dosage or ideal regimen in such cases [12]. It is also possible to proceed to surgical treatment, in which laparoscopic techniques can be used to perform salpingostomy and/or salpingectomy or laparotomy followed by bilateral salpingectomy (usually the choice in low income environments) [3]. There are reported cases of intraoperative diagnosis of bilateral ectopic pregnancy in which salpingectomy of one tube was followed by contralateral tube milking [8] and other reports in which salpingectomy of one tube was followed by methotrexate administration to treat the contralateral fallopian tube [12]. In these two last cases, although the contralateral tube also had an ectopic pregnancy, they were not very compromised, so it was possible to try to preserve them.

Despite many options, the choice of treatment is a dilemma. If on the one hand salpingostomy or milking preserve the tube, on the other hand they increase the chance of a new ectopic pregnancy and increase the chance of failure in assisted reproductive technologies. Bilateral salpingectomy extinguishes the possibility of a new ectopic pregnancy but takes the patient straight to in vitro fertilization for a new pregnancy.

Conclusion

Based on this case report and literature review, it is possible to conclude that the diagnosis of an evident ectopic pregnancy (whether by clinical features, by ultrasound or seriated serum levels of β -hCG) cannot confidently exclude the possibility of a simultaneous bilateral ectopic pregnancy.

Regarding preoperative tests, it is important to remember that the examiner performing an ultrasound must look carefully to the contralateral adnexa in case an ectopic pregnancy is found. In any way, high risk patients with a diagnosed unilateral ectopic pregnancy must be oriented to the possibility of bilateral ectopic pregnancy and conducts must be discussed with the patient before the surgery.

Due to the fact that most bilateral ectopic pregnancies are diagnosed during surgery, we can conclude that it is compulsory and of the utmost importance that both fallopian tubes be routinely carefully examined, independently of the surgical technique chosen, even in the presence of significant adhesions.

The type of treatment chosen is still a dilemma and factors such as patient stability and maintenance of fertility must be considered in the decision making.

Competing Interests

The authors declare that they have no competing interests.

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