CASE REPORT

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Unilateral Tubal Twin Ectopic Pregnancy: A Rare Case Report and Literature Review

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ABSTRACT

Ectopic pregnancy is a common and potentially fatal gynecologic emergency that endangers the mother's life if left untreated and undetected. We present a rare case of a spontaneously conceived unilateral tubal twin ectopic pregnancy. The diagnosis was made using transvaginal ultrasound, and the case was referred to the early pregnancy unit.

Ectopic pregnancy (EP) is a common and life-threatening gynecologic emergency. Prompt surgical intervention is crucial to save the mother's life. Failure to recognize this serious condition carries a high rate of mortality or morbidity. EP ruptures are the leading cause of maternal mortality within the first trimester of pregnancy, with a rate of 9–14% and an incidence of 5–10% of all pregnancy-related deaths.[1] Live twin EPs occur at a frequency of 1 in 125 000, with the first case reported by De Ott.[2] However, the detection of EP has significantly improved in recent years due to the use of a combination of serum quantitative human chorionic gonadotrophin (hCG), transvaginal ultrasound, and laparoscopy.[3]

The detection rate with endovaginal ultrasonography has been reported to be between 60% and 83%.[4] Most EPs occur in the fallopian tube, and twinning accounts for approximately 1.5% of all pregnancies. Furthermore, unilateral ectopic twin pregnancy is a rare condition.[5]

Fishback proposed criteria for the diagnosis of bilateral tubal EP, which required a description of the fetuses or any portion of them, as well as a description of placental material. Norris revised and expanded these criteria, stating that the presence of chorionic villi in each tube is sufficient evidence for the diagnosis.[6] There are three possible explanations for bilateral EP: 1) simultaneous multiple ovulations, 2) sequential impregnation, or 3) transperitoneal migration of trophoblastic cells from one extrauterine pregnancy to the other tube with implantation there.[7]

Risk factors for EP include pelvic inflammatory disease, intrauterine device use, tubal surgery, and assisted reproductive techniques (ARTs). However, in some cases, the etiology remains unknown.[8] The most recognized symptoms are pelvic pain, an adnexal mass, and uterine bleeding.

The current gold standard for the medical care of EPs includes intramuscular methotrexate injections. Intramuscular methotrexate, a folate antagonist, prevents rapid cell division, thus terminating the EP. However, hemodynamic instability, anemia, leukopenia, thrombocytopenia, pelvic discomfort, and hemoperitoneum are contraindications for medical therapy.[9] Furthermore, salpingectomy is the preferred surgical intervention over salpingostomy in cases where the contralateral tube is healthy to reduce the recurrence risk of EP.[2]

CASE REPORT

A 27-year-old Saudi female (gravida 5, para 3 + 1) with an unknown last menstrual period presented to the emergency room with a three-day history of lower left abdominal pain, which worsened a few hours prior to presentation.

There was no history of vaginal bleeding, fever, diarrhea, or vomiting. She had been evaluated at another private hospital, where a diagnosis of missed abortion was considered. She was discharged with

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a narcotic prescription, which partially relieved her pain but did not provide complete relief.

Her medical history revealed a previous laparotomy and right salpingostomy eight years prior for the right tubal EP. She also had a history of three cesarean sections but did not undergo ARTs or take any medications.

On physical examination, the patient was afebrile, and her vital signs were stable. Abdominal palpation revealed tenderness in the lower left quadrant. A pelvic examination showed a normal vagina, a closed cervical os, uterine tenderness, and a palpable left adnexal mass.

Diagnostic testing showed a serum β-hCG level of 17 565 mIU/mL, a hemoglobin level of 10.9 g/dL, a hematocrit of 34%, and normal liver, renal, and electrolyte levels.

A transvaginal ultrasound examination revealed a left adnexal mass measuring 10 × 10 × 8 cm, containing a nonviable twin pregnancy with free fluid surrounding it. Each fetal crown-rump length measured 13.4 mm, corresponding to a gestational age of seven weeks and four days. There was a separating linear echo in the mass between the two fetuses, and no cardiac activity was detected [Figure 1].

An urgent exploratory laparotomy was performed through a Pfannenstiel incision, revealing a hemoperitoneum containing 300 mL and a 10 cm left-sided pelvic mass. The left tubal EP was removed by salpingectomy. The patient was discharged from the hospital on postoperative day 2 and was followed up weekly until her serial β-hCG levels normalized to ensure no remaining pregnancy. Histopathology examination confirmed a fallopian tube twin pregnancy and products of conception. No other abnormalities were found.

DISCUSSION

The incidence of EP has increased since the introduction of ART and tuboplasty. Approximately, one in every 100 pregnancies is ectopic, with the fallopian tube being the most common site (95%), followed by ovarian (< 3%) and other locations such as abdominal, cervical, or cornu (< 1%).[9] While rare, the incidence of twin EPs is estimated to be one in every 20 000 spontaneous pregnancies, with tubal ectopics accounting for one in every 200 EPs.[10] In recent years, the incidence of heterotopic pregnancy associated with in vitro fertilization

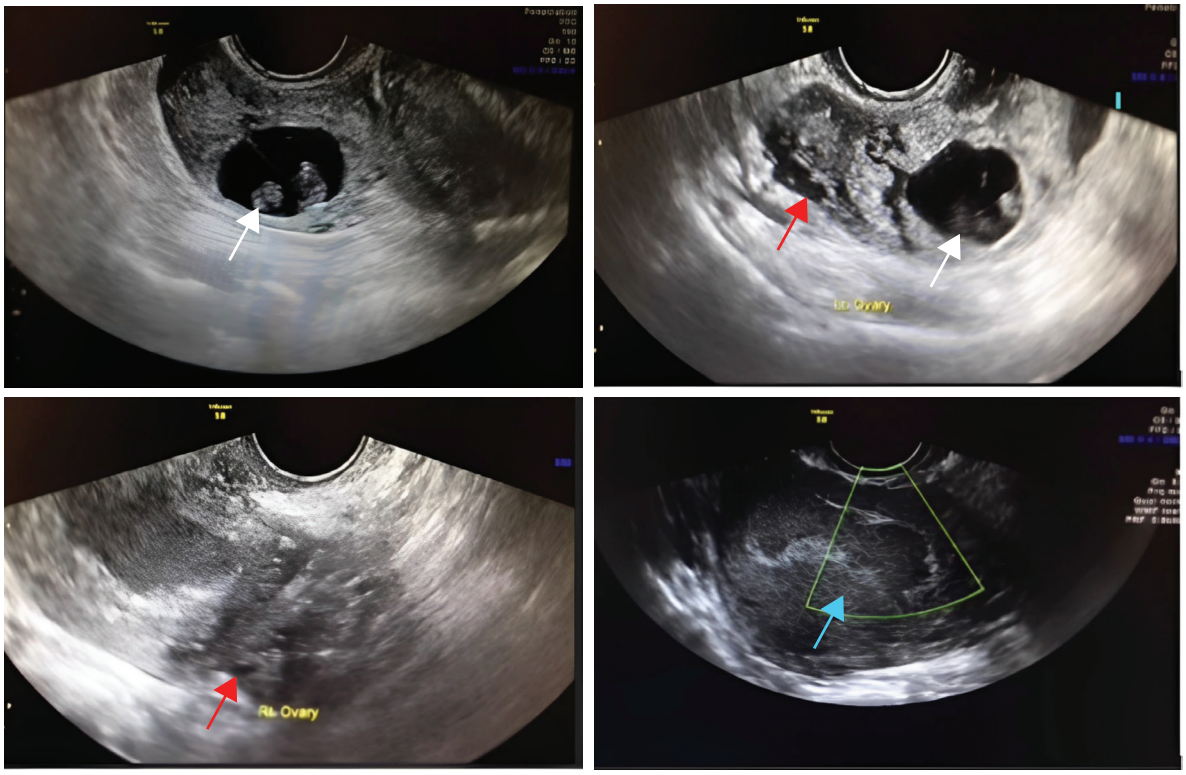

Figure 1: Transvaginal ultrasound depicts a left tubal twin pregnancy attached to the left ovary (white and red arrows, respectively). Doppler echo shows an absent fetal heart and surrounding free fluid (blue arrow).

and embryo transfer has risen to 1–3% of achieved pregnancies.[11]

The diagnosis of EP can be challenging for both expert and non-expert obstetricians. However, maintaining a high index of suspension can aid in reaching a diagnosis, especially in women at high risk for EP. Advances in diagnostic techniques have contributed to the increased recognition of EP cases. Although the number of unilateral twin EPs remains rare, the improvements in detection have led to more cases being reported.[3,9]

The classic triad of EP symptoms includes nonspecific lower abdominal pain, vaginal bleeding, and the presence of an adnexal mass. However, these symptoms were seen in as few as 45% of EP cases.[6] Despite the increasing incidence and morbidity rates, the maternal mortality rate has significantly decreased by 70–90% in developed countries, likely due to early recognition using transvaginal ultrasonography and highly sensitive serum β-hCG tests in suspected cases.[4]

De Ott described the first unilateral twin pregnancy in 1891.[2] Live twin EPs are rare, with an incidence of one in 125 000.[5] Approximately 100 twin EPs have been reported to date, with < 10 cases of unilateral ectopic twin pregnancies, with beating hearts in both embryos reported. Gualandi et al,[12] reported the first case of unilateral tubal pregnancy with cardiac activity in 1994.

Distinguishing between twin and singleton EP can be done through β-hCG levels, which tend to be higher in twin tubal pregnancies compared to singleton tubal pregnancies. Transvaginal ultrasonography is considered the gold standard for differentiation.[3,9] While unilateral twin pregnancy can occur spontaneously, some cases have been reported following in vitro fertilization , detection of the zygote, mechanical obstruction within the tube, hormonal disturbances, smoking, infection (particularly Chlamydia trachomatis), advanced maternal age, adhesions, and previous operations in the contralateral tube, as seen in our case.[13]

Treatment for EP can be conservative or involve surgical interventions. In cases of heterotopic EP, the aim is to be minimally invasive to preserve the development of the intrauterine pregnancy to term, which has been reported in the largest center series at up to 50%. However, in life-threatening conditions such as hemoperitoneum and hemorrhagic shock, laparotomy is indicated. Laparoscopic treatment has shown favorable outcomes in 62.5% of cases,[9,14] and hysteroscopic surgery alone can be considered a valid conservative approach, even in the management of twin cesarean section EP, especially when detected early.[13] Ghanbarzadeh et al,[14] reported a similar case of spontaneous unilateral live tubal twin pregnancy in a patient with a history of previous EP and previous tubal surgery who underwent salpingectomy as the only way of saving her.

While the morbidity and mortality associated with singleton EPs have significantly decreased, the risk of rupture is higher in twin EPs, with rupture occurring in approximately 30–50% of cases. Surgical intervention remains the cornerstone of treatment for these pregnancies.

CONCLUSION

EP is a common and potentially life-threatening gynecologic emergency. The fallopian tube is the most common site for EP. However, if left untreated, tubal twin EP can pose serious health risks and lead to potentially fatal complications. Due to the possible mortality and morbidity, the possibility of twin EP must be taken into consideration based on physical examination and presence of risk factors, which should be carefully assessed through ultrasound scanning. The management strategy for spontaneous unilateral tubal twin pregnancies is not well established due to their rarity. Both laparotomy and laparoscopic salpingectomy have been successfully used in situations similar to our case.

Disclosure

The authors declare no conflicts of interest. Written consent was taken from the patient.

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