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

Gehan Mohammed Atef[1]*, Maryam A Al-Attas[2], Hjazia S Alnaqla[2] and Ibtesam F Alshammari[1]

[1]Department of Obstetrics and Gynecology, King Saud Medical City, Riyadh, Saudi Arabia

[2]College of Medicine, Alfaisal University, Riyadh, Saudi Arabia

Received: 17 April 2023

Accepted: 28 May 2023

*Corresponding author: jehan.attef@yahoo.com

DOI 10.5001/omj.2024.36

Abstract

Ectopic pregnancy is a common and potentially fatal gynecologic emergency that, if left untreated and undetected, endangers the mother's life. Here, we report a rare case of a spontaneously conceived unilateral tubal twin ectopic pregnancy. A transvaginal ultrasound was used to diagnose this case, which was brought to the attention of the early pregnancy unit.

Keyword: Ectopic pregnancy, Unilateral twin ectopic, Salpingostomy. Cesarean section.

Introduction

Ectopic pregnancy (EP) is a common and potentially life-threatening gynecologic emergency. Prompt surgical intervention is crucial to saving the mother's life**.** Failure to recognize such a grave problem carries a high rate of mortality or morbidity. Ectopic pregnancy (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 ectopic pregnancies occur at a frequency of 1 in 125,000, with the first case reported by DeOtt[2]. Although detection of ectopic pregnancy has been raised dramatically in the last few 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 (EVS) has been reported to be between 60 and 83%[4]. Most ectopic pregnancies occur in the fallopian tube, and twinning accounts for approximately 1.5% of all pregnancies; moreover, unilateral ectopic twin pregnancy is a rare entity[5].

Fishback proposed criteria for the diagnosis of bilateral tubal ectopic pregnancy. The criteria 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, making the presence of chorionic villi in each tube sufficient evidence for the diagnosis[6]. There are three possible explanations for a bilateral ectopic pregnancy: 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 ectopic pregnancy include pelvic inflammatory disease (PID), intrauterine device (IUD) use, tubal surgery, and assisted reproductive techniques. However, in some cases, the etiology is 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 (MTX) injections. A folate antagonist called MTX prevents rapid cell division, which terminates EP. Moreover, hemodynamic instability, anemia, leukopenia, thrombocytopenia, pelvic discomfort, and hemoperitoneum are all contraindications for medicinal therapy.[9] Moreover, salpingectomy is the preferred surgical intervention over salpingostomy in the case of a healthy contralateral tube to reduce the recurrence risk of ectopic pregnancy[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 3-day history of lower left abdominal pain that was progressive in course and worsening a few hours prior to the presentation.

There was no history of vaginal bleeding, fever, diarrhea, or vomiting. She was evaluated previously at another private hospital, and a diagnosis of missed abortion has been raised; ultimately, she was discharged with a narcotic prescription that has improved her pain but did not completely relieve it.

Her medical history revealed prior surgery with laparotomy and right salpingostomy for the right tubal ectopic pregnancy eight years ago and a history of three cesarean sections, but she had not been involved in assisted reproductive techniques or given a history of taking medications.

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

Diagnostic testing showed a serum beta-human chorionic gonadotrophin (β-hCG) level of 17565mIU/ml, HGB of 10.9 g/dl, a hematocrit of 34%, and normal liver, renal, and electrolytes.

A transvaginal ultrasound examination revealed a 10 × 10 × 8 cm left adnexal mass containing a nonviable twin pregnancy with free fluid surrounding it. Moreover, each fetal crown-rump length is 13.4 mm, corresponding to a gestational age of 7 weeks and 4 days. A separating linear echo was seen in the mass between the two fetuses, and cardiac activity was absent. Figure1. a, b, c, and d.

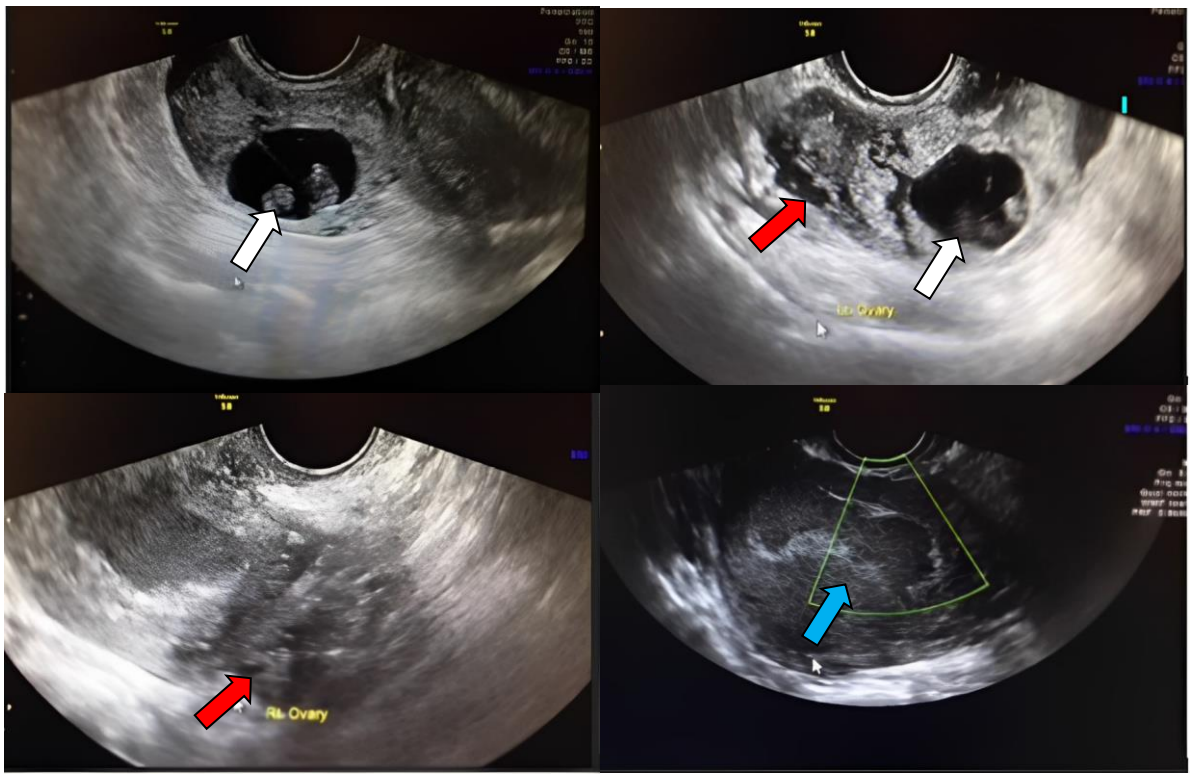

Figure 1: a through d: Transvaginal ultrasound depicts a left tubal twin pregnancy attached to the left ovary –white, red arrows respectively. Doppler Echo showing an absent fetal heart and surrounding free fluid– blue arrow.

An urgent exploratory laparotomy performed via Pfannenstiel incision revealed a hemoperitoneum containing 300 mL and a 10 cm left-sided pelvic mass, and 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 level was normalized to ensure there was no remaining pregnancy. Histopathology examination confirmed a fallopian tube twin pregnancy and products of conception. No other abnormalities were found.

Discussion

The incidence of ectopic pregnancy has increased since the introduction of assisted reproductive techniques (ART) and tuboplasty. As many as one in every 100 pregnancies is an ectopic pregnancy[9]. The commonest site for ectopic pregnancy is the fallopian tube (approximately 95%), with approximately 3% being ovarian in location and the rest (<1%) being abdominal, cervical, or in the cornua. Although quite rare, the incidence of twin ectopic pregnancies is estimated to be 1 in every 20 000 spontaneous pregnancies, and twin tubal ectopics account for 1 in every 200 ectopic pregnancies[10]. In recent years, the incidence of heterotopic pregnancy associated with in vitro fertilization and embryo transfer (IVF-ET) has risen to 1%–3% of achieved pregnancies[11].

The diagnosis of ectopic pregnancy is a daunting task facing expert and non-expert obstetricians alike; however, a high index of suspension may aid in reaching the diagnosis, especially in women at high risk of EP. Recognizing cases of EP has increased recently due to the advances that have been made in the last few years; furthermore, the number of unilateral twin ectopic pregnancies remains rare, with an incidence of 1 in 20,000 spontaneous pregnancies[3, 9.]

Nonspecific lower abdominal pain, vaginal bleeding, and the presence of an adnexal mass are the classic triad of ectopic pregnancy, although no specificity of these symptoms may be seen in as few as 45% of ectopic pregnancy cases[6]. Despite an increase in the incidence and persisting morbidity rate in the last few years, the maternal mortality rate has decreased dramatically by 70–90% in developed countries. This is probably attributed to early recognition using transvaginal ultrasonography and highly sensitive serum beta-HCG tests in suspected cases[4].

In 1891, the first case of unilateral twin pregnancy was described by De Ott. Live twin ectopic pregnancies are rare, with an incidence of one in 1,25,000. 5 Approximately 100 twin ectopic pregnancies have been reported to date. There are fewer than 10 unilateral ectopic twin pregnancies reported with beating hearts in both embryos. Furthermore, Gualandi et al. reported the first case of unilateral tubal pregnancy with cardiac activity in 1994 [12].

The question that has been raised by some experts in the field of gynecology is, "How to differentiate between twin and singleton ectopic pregnancy?" The answer is by the β-hCG level, which will be higher in the case of twin tubal pregnancies than that of a singleton tubal pregnancy. The second way of differentiating is by the gold standard test, which is transvaginal ultrasonography[3, 9.] A unilateral twin pregnancy can occur spontaneously; however, some cases have been reported following in vitro fertilization, detection of the zygote, mechanical obstruction within the tube, hormonal disturbances, active and passive cigarette and tobacco smoking, infection (particularly Chlamydia trachomatis), advanced maternal age, adhesions, the presence of intratubal adhesions, or previous operations in the contralateral tube, as has been mentioned in our case[13].

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

Although the morbidity and mortality linked to singleton ectopic pregnancies have significantly decreased, the risk of rupture is higher in twin ectopic pregnancies, and rupture is thought to happen in 30–50% of cases. The cornerstone of treatment for these pregnancies is still surgical intervention. Furthermore, due to the increased likelihood of treatment failure and rupture with medical care, guidelines advise salpingectomy in the case of ectopic pregnancy with β-hCG greater than 5000 IU/L[1].

Conclusion

Ectopic pregnancy (EP) is a common and potentially life-threatening gynecologic emergency. The commonest site for ectopic pregnancy is the fallopian tube. However, if left untreated, the tubal twin EP is a serious health risk and could result in potentially fatal complications. Due to the possible mortality and morbidity, twin EP must therefore be taken into consideration based on physical examination and the presence of risk factors, which should be carefully checked for on ultrasound scanning. There is no agreement on the appropriate management strategy due to the rarity of spontaneous unilateral tubal twin pregnancies. Laparotomy and laparoscopic salpingectomy have been effectively used in situations comparable to ours.

Competing interests

The authors declared that they have no competing interests. Written consent was taken from the patient.

References

1. Martin A, Balachandar K, Bland P. Management of a spontaneously conceived live unilateral twin ectopic pregnancy in Australia: A case report. *Case Reports Women's Heal*; 30. Epub ahead of print 1 April 2021. DOI: 10.1016/J.CRWH.2021.E00300.

2. Longoria TC, Stephenson ML, Speir VJ. Live unilateral twin ectopic pregnancy in a fallopian tube remnant after previous ipsilateral salpingectomy. *J Clin Ultrasound* 2014; 42: 169–171.

3. Ash KM, Lyons EA, Levi CS, et al. Endovaginal sonographic diagnosis of ectopic twin gestation. *J Ultrasound Med* 1991; 10: 497–500.

4. Hois EL, Hibbeln JF, Sclamberg JS. Spontaneous twin tubal ectopic gestation. *J Clin Ultrasound* 2006; 34: 352–355.

5. Ryan MT, Saldana B. Bilateral Tubal Ectopic Pregnancy: A Tale of Caution. *Acad Emerg Med* 2000; 7: 1160–1163.

6. Amine BHH, Haythem S. Extra-uterine twin pregnancy: case report of spontaneous bilateral tubal ectopic pregnancy. *PAMJ 2015; 20:435* 2015; 20: 1–4.

7. Goswami D, Agrawal N, Arora V. Twin tubal pregnancy: A large unruptured ectopic pregnancy. *J Obstet Gynaecol Res* 2015; 41: 1820–1822.

8. Monochorionic diamniotic twin cervical ectopic pregnancy following assisted conception: a case report - PubMed, https://pubmed.ncbi.nlm.nih.gov/24050036/ (accessed 18 May 2023).

9. Mullany K, Minneci M, Monjazeb R, et al. Overview of ectopic pregnancy diagnosis, management, and innovation. *Womens Health (Lond Engl)*; 19. Epub ahead of print 1 January 2023. DOI: 10.1177/17455057231160349.

10. Lategan H, Gillispie V. Spontaneous Unilateral Tubal Twin Ectopic Pregnancy. *Ochsner J* 2019; 19: 178.

11. Göker ENT, Tavmergen E, Özçakir HT, et al. Unilateral Ectopic Twin Pregnancy Following an IVF Cycle. *J Obstet Gynaecol Res* 2001; 27: 213–215.

12. George M, Nadarajah S, Radiol CO-J coll, et al. Unilateral twin ectopic pregnancy. *hkjr.orgPaperpile* 2010; 12: 186–195.

13. Mollo A, Alviggi C, Conforti A, et al. Intact removal of spontaneous twin ectopic Caesarean scar pregnancy by office hysteroscopy: case report and literature review. *Reprod Biomed Online* 2014; 29: 530–533.

14. Ghanbarzadeh N, Nadjafi-Semnani M, Nadjafi-Semnani A, et al. Unilateral twin tubal ectopic pregnancy in a patient following tubal surgery. *J Res Med Sci* 2015; 20: 196.