Spontaneous unilateral tubal twin ectopic pregnancy: Case report and literature review

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ABSTRACT

Ectopic pregnancy is defined as implantation of a pregnancy outside the uterine cavity. Its twin form is exceptionally rare and potentially life-threatening, making early diagnosis essential. We report the case of a 30-year-old primigravida diagnosed with a spontaneous unilateral tubal twin ectopic pregnancy, successfully treated with right laparoscopic salpingectomy without complications. Although uncommon, twin ectopic pregnancy should be considered in patients presenting with abnormally elevated β-hCG levels and compatible ultrasound findings, as prompt recognition is critical to reduce maternal morbidity.

KEYWORDS

Twin ectopic pregnancy, spontaneous pregnancy, unilateral.

Introduction

Ectopic pregnancy (EP) is defined as a pregnancy that occurs outside the uterine cavity, most commonly in the ampullary portion of the fallopian tube [1]. Twin ectopic pregnancy (TEP) is exceedingly rare, particularly when it occurs spontaneously and both embryos are implanted within the same tube. Because the risk of tubal rupture is higher than in singleton EP, early diagnosis is critical. Owing to its exceptional rarity, no specific management or treatment guidelines have been established.

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Case report

A 30-year-old primigravida presented to the Emergency Department with vaginal bleeding, hypogastric pain, and 7 weeks of amenorrhea. She reported no relevant medical or surgical history and no additional symptoms. On physical examination, hypogastric tenderness was noted on deep abdominal palpation, without signs of peritoneal irritation. Vital signs were stable. Pelvic examination revealed a closed cervix and tenderness in the right adnexal region.

Laboratory testing showed a serum β-subunit of human chorionic gonadotropin (β-hCG) level of 56,172 mIU/mL. All other laboratory parameters were within normal limits. Transvaginal ultrasound demonstrated an empty uterus with an endometrial thickness of 9 mm. A 40 × 38 mm anechoic image, consistent with a corpus luteum, was identified in the left ovary, while the right ovary had a normal ultrasound appearance. Adjacent to the right ovary, two gestational sacs were visualized, each containing an embryo with a crown–rump length of 8.9 mm and 8.8 mm, corresponding to 6 weeks and 6 days of gestation. Cardiac activity was detected in both embryos (Figure 1).

Figure 1. Transvaginal ultrasound revealing two embryos with heartbeat within the right fallopian tube.

The patient was diagnosed with a unilateral tubal twin pregnancy. The findings and available therapeutic options were discussed, and laparoscopic surgical management was selected. The procedure was performed using a 12-mm umbilical trocar and two 5-mm accessory trocars placed in the left iliac region.

Intraoperatively, a right adnexal mass was identified, corresponding to a tubal lesion measuring approximately 84 × 45 mm (Figure 2). After careful inspection of the entire abdominal cavity, a right salpingectomy was performed (Figures 3 and 4) without complications. The surgical specimen was retrieved in a closed extraction bag through the umbilical trocar and sent for histopathological examination. The postoperative course was uneventful, and the patient was discharged 12 hours after surgery. Histopathological analysis confirmed a tubal TEP, identifying two embryos.

Figure 2. Right Fallopian tube.

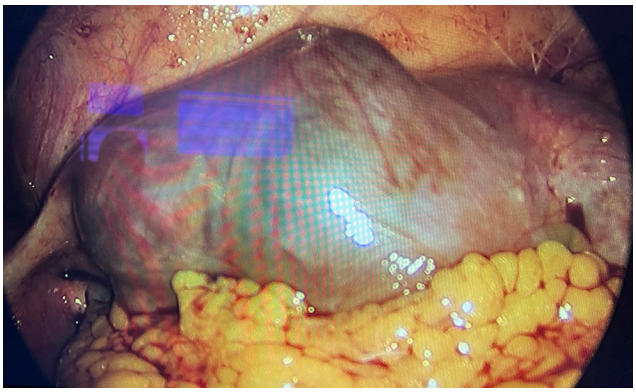

Figure 3. Surgical piece of the right salpingectomy.

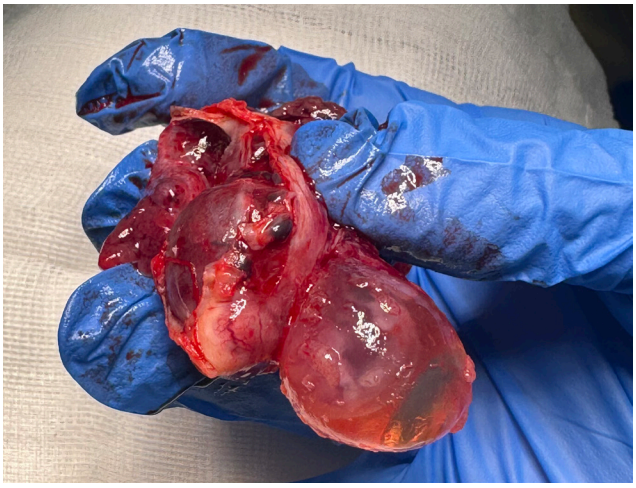

Figure 4. Surgical piece opened where two embryos are visualized.

Discussion

A literature review was conducted using a predefined search strategy. The databases PubMed/MEDLINE, Scopus, Embase, and ScienceDirect were searched using the following terms: ("unilateral tubal twin ectopic pregnancy" OR "unilateral twin tubal pregnancy" OR "spontaneous live unilateral tubal twin ectopic pregnancy") NOT (heterotopic), without time restrictions. Clinical case reports, case series, and review articles were included.
A total of 82 articles were initially identified. After duplicate removal using Zotero Reduplicator and additional manual screening, 45 articles met the eligibility criteria. The main clinical characteristics extracted from these studies are summarized in Tables 1, 2, 3, and 4, categorized according to therapeutic approach: medical management, laparoscopy, laparotomy, or unspecified treatment.

The incidence of EP has increased in recent years, largely due to the growing prevalence of risk factors such as pelvic inflammatory disease, prior pelvic surgery, and the use of assisted reproductive techniques [3]. Tubal implantation remains the most common location, accounting for more than 90% of cases [23]. In contrast, TEP are exceptionally rare, with an estimated incidence of approximately 1 in 125,000 spontaneous pregnancies [1]. The presence of fetal cardiac activity in both embryos represents an even more unusual clinical finding.

TEP are associated with higher morbidity and mortality than singleton EP, particularly when both embryos are located within the same fallopian tube. The increased tubal distension results in a substantially higher risk of rupture, reported in 30–50% of cases [2].

The first documented description of a unilateral TEP dates back to 1891 [4]. Nearly a century later, the introduction of ultrasonography made preoperative diagnosis of this rare condition possible [7].

Most reported cases involve young women, with a mean age of 31.9 ± 5.99 years (range 24–51 years). Among the published cases, identifiable risk factors were present in 22 of 46 patients (47.8%). The most frequently reported risk factors included a history of pelvic surgery (notably cesarean section), previous EP, pelvic inflammatory disease, and the use of assisted reproductive techniques.

Abdominal pain was the most common presenting symptom, reported in all cases, followed by vaginal bleeding. Owing to the larger size of TEP, symptoms and complications—particularly tubal rupture—tend to occur earlier, leading to diagnosis most frequently after 6–7 weeks of amenorrhea. In this series, the latest diagnosis was reported at 12 weeks [33].

Serum β-hCG levels were generally higher than expected for the corresponding gestational age. However, this parameter showed wide interindividual variability and should therefore be interpreted with caution [47,48].

Transvaginal ultrasound was the most useful imaging modality for determining the location of the pregnancy, with a reported

Table 1. Record of previous published cases: main clinical features treated with methotrexate. IVF: In Vitro Fertilization. CRL: Crown-Rump Length.

Author	Year	Age	Risk factors	Symptoms	β-hCG mIU/mL	Ultrasound	Treatment
Fernandez et al. [6]	1993	32	Ectopic pregnancy	Abdominal pain and vaginal bleeding	3,640	Two embryos CRL 11 y 13 mm; negative cardiac activity; unruptured	Methotrexate two doses (1st intrasacular 2nd IM)
Karadeniz et al. [19]	2008	26	IVF; smoker	Abdominal pain and vaginal bleeding	763	Two gestational sacs; unruptured.	Methotrexate multiple doses (days 1, 3, 5, 7).
Arikan et al. [22]	2010	26	None	Abdominal pain and vaginal bleeding	18,780	Two embryos CRL 11.2 and 7.9 mm; negative cardiac activity; unruptured	Methotrexate IM single dose
Betti et al. [32]	2018	31	Endometriosis; ectopic pregnancy (salpingectomy)	-	13,217	Adnexal mass 29 x 16mm; unruptured	Methotrexate 3 doses (days 1, 7, 14), after which rupture occurred: Laparoscopic salpingectomy was then performed
Madaan et al. [39]	2021	32	C-section; ectopic pregnancy	Abdominal pain and vaginal bleeding	10,000	Two embryos CRL 18 and 20 mm; positive cardiac activity; unruptured	Methotrexate single dose intrasacular
Forbes et al. [46]	2024	24	3 C-section; C. trachomatis infection; smoker	Abdominal pain and vaginal bleeding	798	Two gestational sacs; unruptured.	Methotrexate IM two doses (days 1 and 4)

sensitivity of 87–99% and specificity of 94–99% [2]. Nevertheless, visualization of two embryos was not always possible, and in some cases the diagnosis of unilateral TEP was established only postoperatively through histopathological examination of the surgical specimen [34]. The combined use of transvaginal ultrasound and β-hCG measurement has substantially improved the diagnostic accuracy for EP, even in rare presentations such as TEP.

This review did not identify any preference for right- or left-sided tubal involvement, with both sides reported at similar frequencies.

Regarding management, because of the extremely low incidence of spontaneous unilateral tubal TEP, no standardized therapeutic protocol exists [49]. The choice of treatment approach was mainly determined by the patient's hemodynamic stability, prior surgical history, and/or surgeon preference.

Overall, surgical management was far more common than medical treatment (40 cases vs. 5 cases in this series). Salpingectomy was the most frequently performed procedure, via either laparotomy or laparoscopy. Laparotomy was used in 17 cases, while laparoscopy was performed in 23 cases (one case was not specified). Despite the increasing trend toward minimally invasive surgery, laparotomy remains the preferred approach in cases of tubal rupture, hemodynamic instability, or based on the surgeon's judgment.

Although alternative surgical techniques have been successfully used for singleton EP, their effectiveness in TEP has not been adequately studied.

In hemodynamically stable patients without medical comorbidities that contraindicate methotrexate (MTX), such as liver or kidney disease [3], and with no signs of tubal rupture, medical management with MTX has been attempted. In the present series, six cases were treated with MTX; however, one of these subsequently required surgical intervention due to tubal rupture [32].

Overall prognosis depends largely on early diagnosis and prompt management, with most reported cases showing favorable maternal outcomes. The principal risk remains tubal rupture with severe hemorrhage. Therefore, it is essential to recognize the increased risk associated with suspected unilateral tubal TEP and to manage these cases proactively and appropriately.

Highlights

- TEP is a rare condition, but one that must be considered due to the higher risk of rupture.
- A warning sign of a TEP is the finding of unexpectedly high β-hCG levels.
- Difficulty in identifying both embryos through ultrasound may occur.
- There are no standardized treatment recommendations; surgical approach is most commonly chosen.

Conclusion

Spontaneous unilateral tubal TEP is an extremely rare condition and may be associated with significant maternal morbidity if not promptly recognized and treated. It should be suspected in the presence of pelvic pain, abnormally elevated β-hCG levels, and suggestive ultrasound findings, even in the absence of classic risk factors for EP. Management should be individualized, with surgical treatment remaining the preferred approach, although selected cases have been successfully managed medically. The development of evidence-based guidelines derived from the existing scientific literature is essential to improve diagnosis, management, and outcomes in these rare cases.

Table 2. Record of previous published cases: main clinical features treated with laparoscopic approach.

Author	Year	Age	Risk factors	Symptoms	β-hCG mIU/mL	Ultrasound	Treatment
Gualandi et al. [7]	1994	-	-	-	-	Two embryos; positive cardiac activity	Laparoscopic salpingectomy
Parker et al. [8]	1999	27	Inflammatory pelvis disease; bilateral tubal obstruction	Abdominal pain and vaginal bleeding	1,855	Two embryos; positive cardiac activity; unruptured	Laparoscopic salpingectomy
Göker et al. [10]	2001	37	IVF	Vaginal bleeding	55,845	Two embryos; positive cardiac activity; unruptured	Laparoscopic salpingectomy
Sur et al. [14]	2005	24	C. trachomatis infection.	Abdominal pain and vaginal bleeding	10,500	Two embryos; positive cardiac activity; ruptured	Laparoscopic salpingectomy
Eddib et al. [15]	2006	41	None	Abdominal pain	31,672	Two embryos; positive cardiac activity; ruptured	Laparoscopic salpingectomy
Dede et al. [16]	2008	24	None	Abdominal pain and vaginal bleeding	1,200	Adnexal mass 4 cm; unruptured	Laparoscopic salpingostomy
Summa et al. [18]	2008	31	None	-	22,447	Two embryos CRL 4 mm; positive cardiac activity	Laparoscopic salpingectomy
Karanjgaokar et al. [20]	2009	36	Curettage	Abdominal pain and vaginal bleeding	38,900	Two embryos CRL 11 and 6 mm; positive cardiac activity; ruptured	Laparoscopic salpingectomy
Luengo et al. [21]	2009	35	None	Abdominal pain and vaginal bleeding	52,382	Two embryos CRL 8.6 and 9.9 mm; positive cardiac activity; ruptured	Laparoscopic salpingectomy
Gallo del Valle et al. [23]	2010	24	None	Nausea	-	Two embryos; positive cardiac activity; unruptured	Laparoscopic salpingectomy
George et al. [24]	2010	38	IVF; inflammatory pelvic disease	-	36,796	One sac with embryo CRL 8 mm with positive cardiac activity and another sac without embryo; unruptured.	Laparoscopic salpingectomy
Longoria et al. [26]	2013	44	Ectopic pregnancy; C. trachomatis infection	-	21,989	Two embryos according 7+2 weeks; positive cardiac activity; unruptured	Laparoscopic salpingectomy (removal of remnant fallopian tube).
Vohra et al. [1]	2014	34	Pelvic inflammatory disease; smoker	Abdominal pain and vaginal bleeding	6,927	Two embryos CRL 1.5 and 1.6 mm; negative cardiac activity; unruptured.	Laparoscopic salpingectomy
Yamane et al. [29]	2015	27	None	Vaginal bleeding	4,650	Two embryos CRL 12 and 7 mm; negative cardiac activity; unruptured	Laparoscopic salpingectomy
Kim et al. [30]	2018	31	None	Abdominal pain and nausea	35,672	Two embryos CRL 8.3 and 7.8 mm; positive cardiac activity; ruptured	Laparoscopic salpingectomy
Felemban et al. [31]	2018	30	Ectopic pregnancy	-	4,960	Two embryos one positive cardiac activity and another negative cardiac activity; ruptured	Laparoscopic salpingectomy (removal of remnant fallopian tube).
Seak et al. [33]	2019	32	None	Abdominal pain	-	Two embryos CRL 55 mm; positive cardiac activity; unruptured	Laparoscopic salpingectomy
Lategan et al. [35]	2019	40	C-section	Abdominal pain and vaginal bleeding	23,359	Two embryos; negative cardiac activity; unruptured	Laparoscopic salpingostomy
El Moussaoui et al. [36]	2020	28		Vaginal bleeding	3966	Two embryos CRL 3.8 and 3.6 mm; positive cardiac activity; unruptured	Laparoscopic salpingectomy
Pek et al. [37]	2020	31	None	Abdominal pain	24,271	Two embryos CRL 6.4 and 7.9 mm; positive cardiac activity; unruptured	Laparoscopic salpingectomy
Mahajan et al. [38]	2020	40	None	Abdominal pain and vaginal bleeding	28,500	Two embryos; positive cardiac activity; ruptured	Laparoscopic salpingectomy
Martin et al. [40]	2021	36	None	Asymptomatic	11,870	Two embryos CRL 7.7 and 6.9 mm; positive cardiac activity; unruptured	Laparoscopic salpingectomy
Öztürk et al. [44]	2023	31	None	Abdominal pain and vaginal bleeding	25,696	Two embryos CRL 11.2 and 11.4 mm; positive cardiac activity; unruptured	Laparoscopic salpingectomy
Our present case	2025	30	None	Abdominal pain and vaginal bleeding	56,172	Two embryos CRL 8.9 and 8.8 mm; positive cardiac activity; unruptured	Laparoscopic salpingectomy

Table 3. Record of previous published cases: main clinical features treated with laparotomic approach.

Author	Year	Age	Risk factors	Symptoms	β-hCG mIU/mL	Ultrasound	Treatment
De Ott et al. [4]	1891	-	-	-	-	-	Laparotomic salpingectomy
Storch et al. [5]	1976	34	-	Abdominal pain and vaginal bleeding	-	-	Laparotomic salpingo-oophorectomy
Hanchate et al. [9]	2001	38	None	Abdominal pain and nausea	-	Two embryos according 6 weeks; positive cardiac activity; ruptured	Laparotomic salpingectomy
Shivanand et al. [11]	2004	25	-	Abdominal pain	-	Two embryos; positive cardiac activity	Laparotomic salpingectomy
Hois et al. [12]	2005	31	Ectopic pregnancy	Vaginal bleeding	14,000	Two embryos; one positive and another negative cardiac activity; unruptured	Laparotomic salpingectomy
Rolle et al. [13]	2005	24	C. trachomatis, N. gonorrhoeae, Herpes simplex virus 2 and T. pallidum infection	Abdominal pain	263	Two embryos according 9+3 weeks; negative cardiac activity; unruptured	Laparotomic salpingo-oophorectomy
Tam et al. [17]	2008	27	C-section; ectopic pregnancy; endometriosis	Abdominal pain.	3,500	Two embryos, one positive and another negative cardiac activity; unruptured	Laparotomic salpingectomy
Ghike et al. [25]	2011	34	None	Abdominal pain	-	Two gestational sacs; ruptured	Laparotomic salpingectomy
Ghanbarzadeh et al. [27]	2014	31	Tubal surgery; ectopic; pregnancy; curettage	Abdominal pain and vaginal bleeding	1,750	Two embryos CRL 16 and 17 mm; positive cardiac activity; ruptured	Laparotomic salpingectomy
Goswami et al. [28]	2015	25	None	Abdominal pain and vaginal bleeding	10,800	Adnexal mass 6 cm; unruptured	Laparotomic salpingectomy
Samy et al. [34]	2019	32	None	Abdominal pain		Adnexal mass 7 cm; ruptured	Laparotomic salpingectomy
Jain et al. [41]	2021	35	None	Abdominal pain		Two gestational sacs; ruptured	Laparotomic salpingo-oophorectomy
Gure Eticha [42]	2022	30	2 C-sections	Abdominal pain	86,456	Two embryos; positive cardiac activity; unruptured	Laparotomic salpingectomy
Samha et al. [43]	2023	39	C-section	Abdominal pain and vaginal bleeding	63,000	Adnexal mass 3 cm; ruptured	Laparotomic salpingectomy
Atef et al. [2]	2024	27	3 C-sections; ectopic pregnancy	-	17,565	Two embryos CRL 13.4 mm; negative cardiac activity; ruptured	Laparotomic salpingectomy

Table 4. Record of previous published cases: main clinical features with surgical approach (unspecified: laparoscopic vs. laparotomic).

Author	Year	Age	Risk factors	Symptoms	β-hCG mIU/mL	Ultrasound	Treatment
Zhang et al. [45]	2023	51	IVF		2,408	Two gestational sacs; unruptured	Bilateral salpingectomy

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Author contributions

Sandra Borja-Vergel performed the literature review and wrote the manuscript. José Olmedilla-Bahillo and María Martín-Gómez literature review and revision of the manuscript.
Sandra Borja-Vergel, Sonia M. García-García, and Paula Parrondo-Sánchez contributed to patient care, data acquisition and interpretation, and selection of the histopathological images included in the manuscript.
María J. Cancelo-Hidalgo contributed to drafting the manuscript and critically revising the article for important intellectual content.
All authors approved the final submitted manuscript.

Patient Consent

Informed consent was obtained from the patient. Institutional approval was not required for publication of this case report.

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The authors declare no conflict of interest related to this publication.