

Spontaneous Asymptomatic Unilateral Dichorionic-Diamniotic Twin Tubal Pregnancy Diagnosed with Ultrasound: Case Report and Literature Review

Yuan M¹, Jin C², Wang L¹, Gao C¹, Wang X^{3*} and Song Y^{4*}

¹Department of Obstetrics and Gynecology, Jiaozhou Central Hospital of Qingdao, P.R. China

²Department of Cell Biology, Shandong University School of Medicine, P.R. China

³Department of Obstetrics and Gynecology, Shanghai General Hospital, School of Medicine, Shanghai Jiao Tong University, P.R. China

⁴Department of Special Examination, Jiaozhou Central Hospital of Qingdao, P.R. China

*Corresponding author:

Xiaoyun Wang,
Department of Obstetrics and Gynecology,
Shanghai General Hospital, School of Medicine,
Shanghai Jiao Tong University, 650
XinSongjiang Road, Shanghai,
P.R. China 21600
E-mail: littleyun2007@sina.com.

Yanpeng Song,
Department of Special Examination,
Jiaozhou Central Hospital of Qingdao,
29 Xuzhou Road of Jiaozhou, Qingdao,
P.R. China 266300,
E-mail: songyanpeng719@163.com.

Received: 21 Mar 2021

Accepted: 08 Apr 2021

Published: 13 Apr 2021

Copyright:

©2021 Wang X, Song Y. This is an open access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and build upon your work non-commercially.

Citation:

Wang X, Song Y, Spontaneous Asymptomatic Unilateral Dichorionic-Diamniotic Twin Tubal Pregnancy Diagnosed with Ultrasound: Case Report and Literature Review. *Ame J Surg Clin Case Rep.* 2021; 2(8): 1-5.

Abbreviations:

β-hCG: β-Human Chorionic Gonadotropin; MTX: Methotrexate; ART: Assisted Reproductive Technology; IVF: In Vitro Fertilization; MRI: Magnetic Resonance Imaging; lap salpingectomy: Laparoscopic Salpingectomy; paro salpingectomy: Laparotomy Salpingectomy

#Authors Contributed:

Wang X, Song Y. There are equally contributed to this work.

Keywords:

Unilateral Tubal Twin Pregnancy; Transvaginal Ultrasound; Risk Factors; Laparoscopy; Preoperative Diagnosis; Case Report.

1. Abstract

1.1. Background: Spontaneous unilateral tubal twin pregnancy is a rare condition with an incidence of 1 out of 20,000 - 125,000 pregnancies. We here in report a rare case of spontaneous asymptomatic unilateral dichorionic-diamniotic twin tubal pregnancy.

1.2. Case presentation: A 32-year-old Chinese woman, gravida 5, para 2, was referred to the gynecology department of our hospital for complaining of menopause for 40 days without vaginal bleeding and lower abdominal pain. Her serum β-human chorionic gonadotropin (β-hCG) level was 13,414 mIU/mL and ultrasound revealed left tubal twin pregnancy of 6 weeks' gestation. Transvaginal ultrasound and laparoscopy suggested the twins were dichorionic and diamniotic.

1.3. Conclusion: Unilateral tubal twin ectopic pregnancies can be adequately diagnosed with transvaginal ultrasound and serum β-hCG test. Laparoscopic salpingectomy provides successful treatment of unilateral tubal twin gestation with short recovery time.

2. Background

Ectopic pregnancy is referred to a pregnancy that fertilized ovum implants on any site other than the normal uterine cavity. Missed diagnosis of ectopic pregnancy could lead to tubal rupture, hemoperitoneum and hemorrhagic shock. Tubal twin ectopic pregnancy first reported by De Ott in 1891 is still a rare event. Tubal twin pregnancy is a life-threatening complication, early diagnosis and immediate intervention before rupture can save the patient's life. It was difficult in diagnosing unilateral tubal twin pregnancies by ultrasound before surgery 20 years ago [1]. The estimated incidence of tubal twin pregnancy was 1 in 20,000 spontaneous pregnancies [2]. More than 100 tubal twin pregnancy cases have been reported till 2018 [3]. The epidemiology, risk factors and characteristics of unilateral tubal twin pregnancy are not well understood. We describe another case of unilateral dichorionic-diamniotic tubal twin pregnancy, which was detected at 6 weeks' menstrual age through transvaginal ultrasound in a patient without vaginal bleeding and

lower abdomen pain. Furthermore, we also review all cases of unilateral tubal twin pregnancy since 2000 to provide clinical basis for the management of tubal twin pregnancy.

3. Case presentation

A 32-year-old patient (gravida 5, para 2) had a history of a vaginal delivery 8 years earlier and a cesarean section 3 years ago. Due to amenorrhea for 6 weeks, she went to our hospital for a routine examination. She did not have vaginal bleeding or abdominal pain. Her previous menstrual cycles were regular. The transvaginal ultrasound revealed endometrial thickness was 12 mm, no intrauterine gestational sac, no free fluid in the abdomen. In the left adnexa, adjacent to the ovary, a complex mass measuring 29*35mm encompassing to a further evaluation 2 thick-walled cystic masses measuring 10 and 10 mm, with two yolk sacs at the left adnexal region and fetal cardiac activity was negative (Figure 1). Her serum β -hCG was 13,414 mIU/mL. Her vital signs were

stable. There was no abdominal tenderness. On physical examination, just slight left adnexal tenderness was elicited. No vaginal bleeding was detected. Left tubal twin pregnancy was diagnosed so she was admitted to our emergency room. Surgical management was planned due to her no fertility desire and the large size of the adnexal mass. At laparoscopy, there was no hemoperitoneum, the uterus, left ovary and right adnexa were normal. The left fallopian tube was significantly thickened and distended mainly in tubal ampulla, sized 4*3*3 cm. Surprisingly, the left tube was intact with no evidence of any breach in the tubal wall. Left salpingectomy was carried out. The tube contained two embryos in separate gestational sacs (Figure 1). The pathology confirmed unilateral dichorionic-diamniotic twin tubal pregnancy. The postoperative period was uneventful. Her serum β -hCG fell down to normal 3 weeks after surgery. The patient was well on 12-month follow up.



Figure 1: (a-c) Transvaginal ultrasound shows two distinct gestational sacs in the left adnexal region with no fetal heart activity. The uterus is empty. (d) Intraoperative photograph shows the unruptured left tubal ectopic pregnancy with 2 distinct gestational sacs.

4. Discussion

An ectopic pregnancy is a pregnancy occurring any site other than the uterine cavity, and over 98% implant in the fallopian tube. Ectopic pregnancy develops in almost 2% spontaneous pregnancies [4]. In recent years, incidence of ectopic pregnancy has increased due to assisted reproduction technology. Both in developed country and developing country, tubal ectopic pregnancy remains the most common cause of maternal mortality in the first trimester of pregnancy [5]. The prevalence of ectopic pregnancy has increased while the mortality has declined because of increased sensitivity of serum β -hCG immunoassay and improved quality of transvaginal

ultrasound [6]. Unilateral tubal twin pregnancy is extremely rare as it represents only 0.5% of ectopic pregnancies with incidence of 1 in 20,000 pregnancies [2]. The incidence of live tubal twin pregnancy is even rarer, and has been estimated to be 1 out of 125,000 pregnancies [7]. The risk factors for ectopic pregnancy were tubal damage as a result of surgery or infection, smoking, age, prior spontaneous abortions, history of infertility, and previous use of an intrauterine device [5, 8]. There have been more than 100 tubal twin pregnancy cases reported since 1891[3]. Among these cases, most were diagnosed during or after surgery. Till 2006, only 8 cases of unilateral tubal twin pregnancy in the literature had been

diagnosed pre operatively [9]. With the availability of advanced and accurate laboratory methods, improved quality of high-resolution transvaginal ultrasound and awareness in management of ectopic pregnancy, the cases diagnosed before surgery significantly increased and mortality and morbidity due to ectopic pregnancy decreased. The preoperative diagnosis of unilateral multiple gestation is exceedingly difficult to make. It was reported that most cases of unilateral tubal were monozygotic [10]. Few cases of di-

zygotic unilateral tubal twin pregnancy have been described. Foetal heart activities were visualized in few tubal twin pregnancies cases and less than 5 cases had been attempted to treat with methotrexate. In order to analyze and clarify clinical characteristics of unilateral tubal twin pregnancy, we performed a systematic review of associated cases diagnosed with transvaginal ultrasound since 2000. Twenty related cases were found (Table 1 and 2). Here, we describe another unilateral dichorionic-diamniotic tubal twin pregnancy diagnosed with ultrasound.

Table 1: General characteristics of reported unilateral tubal twin pregnancy diagnosed 141 by ultrasound since 2000

Author	Year	Age	GnPn	Fallopian Tube	β-hCG	Gestational week	Fetal size	Fetal heart activity
Goker et al(11)	2001	37	G1P0	Left	55845	6 weeks	6 weeks/6 weeks	visible
Hanchate et al(7)	2002	38	G2P2	Left	NR	2 months	6 weeks/6 weeks	visible
Sur et al(12)	2005	24	G0P0	Left	10500	6 weeks	NR	visible
Urnsak et al(13)	2006	20	G0P0	Right	8200	8 weeks	NR	NR
Karadeniz et al(14)	2008	26	G0P0	Left	763	NR	NR	NR
Tam et(15)	2008	27	G3P1	Right	3500	6 ⁴ weeks	NR	visible/negative
Summa et al(16)	2009	31	G1P0	Right	22477	49 days	4mm/4mm	visible
Svirsky et al(17)	2010	30	G2P2	Left	20700.6	6 weeks	6 ² weeks/6 ² weeks	visible visible
		39	G1P0	Left	3374	6 ² weeks	6 ² weeks/6 ² weeks	
Arikan et al(18)	2011	26	G0P0	Left	18780	7 weeks	7 ² weeks/6 ⁵ weeks	Negative
Fambrini et al(19)	2012	31	G0P0	Left	4847	6 weeks	16*9mm/12*9mm	Negative
Samardzic et al(20)	2014	30	NR	Right	19236	6 weeks	6 weeks/6 weeks	78/113
Longoria et al(21)	2014	44	G5P1	Left	21989	8 weeks	7 ² weeks/7 ² weeks	156/160
Ghanbarzadeh et al(22)	2015	31	G3P1	Right	1750	5 weeks	8 weeks/8 weeks	visible
Kim et al(23)	2018	31	G1P0	Right	35672.3	7.7 weeks	6.7weeks/6.6weeks	visible
Betti et al(3)	2018	31	G2P0	Left	13217	6 ² weeks	5mm /16mm	Negative
Nepal et al(24)	2019	29	NR	Right	348000	9 weeks	NR	Negative
Tsakiridis et al(25)	2019	34	G0P0	Right	4544	6 weeks	NR	Negative
Seak et al(26)	2019	37	G2P1	Right	NR	12 ⁶ weeks	12 ¹ weeks/12 ⁴ weeks	185/180
Lategan et al(27)	2019	40	G1P1	Right	23359	6 weeks	8 ² weeks/7 ⁶ weeks	Negative
Present study	2020	32	G5P2	Left	13414	6 weeks	6 weeks /6 weeks	Negative

Table 2: Clinical symptoms, risk factors, characteristics and the treatment of reported unilateral tubal twin pregnancy diagnosed before surgery since 2000

Author	Clinical symptoms	Risk factors	Treatments	Monozygotic/ dizygotic
Goker et al(11)	vaginal bleeding	infertility (IVF)	lap salpingectomy	NR
Hanchate et al(7)	vaginal bleeding and pain	No	paro salpingectomy	monochorionic
Sur et al(12)	pain and vaginal bleeding	chlamydia infection	lap salpingectomy	dichorionic
Urnsak et al(13)	pain and vaginal bleeding	NR	NR	NR
Karadeniz et al(14)	vaginal bleeding and pain	infertility (ART)	MTX	NR
Tam et(15)	pain	tubal surgery	paro salpingectomy	dichorionic
Summa et al(16)	No	No	lap tube anastomosis	monochorionic
Svirsky et al(17)	suspicion of tubal pregnancy	NR	left salpingectomy	monochorionic
	No	infertility (IVF)	lap salpingectomy	dichorionic
Arikan et al(18)	vaginal bleeding and pain	infertility	MTX	dichorionic
Fambrini et al(19)	pain and vaginal bleeding	NR	lap salpingectomy	dichorionic
Samardzic et al(20)	pain and nausea	smoking	lap salpingectomy	monochorionic
Longoria et al(21)	No	tubal surgery	lap salpingectomy	monochorionic
Ghanbarzadeh et al(22)	pain and vaginal bleeding	tubal surgery	paro salpingectomy	monochorionic
Kim et al(23)	nausea, vomiting, and pain	abortion	right salpingectomy	dichorionic
Betti et al(3)	No	tubal surgery	MTX, lap salpingectomy	NR
Nepal et al(24)	abdominal pain.	infertility(IVF)	lap salpingoophorectomy	dichorionic
Tsakiridis et al(25)	pelvic pain	No	MTX	NR
Seak et al(26)	pain	No	lap salpingectomy	monochorionic
Lategan et al(27)	vaginal bleeding and pain	infertility (IVF)	lap salpingostomy	NR
Present study	No	abortion	lap salpingectomy	dichorionic

The median age of unilateral tubal twin pregnancy is 31, with the youngest case 20-year-old and the oldest case 44-year-old. About 76.19% cases are older than 30-year-old. Among all cases, 31.58% tubal twin pregnancies were the first pregnant. More than half cases (57.89%) were nulliparous. The incidence of left tubal twin pregnancy is almost equal to that of right. Median serum β -hCG is 13414 mIU/mL. Serum β -hCG of nearly 32% unilateral tubal twin pregnancies are less than 5000 mIU/mL while most (63.16%) cases are higher than 10000 mIU/mL. Most unilateral tubal twin pregnancies were diagnosed at gestational age of 6-8 weeks. Surprisingly, a live tubal twin pregnancy was diagnosed at 12 weeks of gestation [26]. Among all cases, fetal heart activity is visualized through transvaginal ultrasound or MRI in 63.16% cases (12/19). Abdominal pain (66.7%, 14/21) is the most common symptom in these cases, and vaginal bleeding (42.9%, 9/21) ranks the second. Not all tubal twin pregnancy is symptomatic. Nearly 23.81% (5/21) cases are asymptomatic. Risk factors of tubal twin pregnancy among these cases are as follows: infertility (28.57%, 6/21), tubal surgery (19.05%, 4/21), abortion (9.52%, 2/21), chlamydia infection (4.76%, 1/21) and smoking (4.76%, 1/21). However, nearly 20% cases have no risk factors. Most (85%) tubal twin pregnancy has been treated surgically. Only 4 cases of tubal twin pregnancies have been attempted to treat by methotrexate, with one failure. The number of cases treated by laparoscopy is 3 times more than that of laparotomy. Nearly 88.24% unilateral tubal twin pregnancies have been treated by salpingectomy compared with 11.76% cases treated by salpingostomy or anastomosis. In this study, we finally found that the incidence of monozygotic tubal twin pregnancy cases is almost the same as that of dizygotic cases, which is different from the previous studies.

5. Conclusion

Unilateral tubal twin pregnancy is a rare ectopic pregnancy with difficulties in early diagnosis. Infertility is one of the most important risk factors of tubal twin pregnancy. Unilateral tubal twin pregnancy could be treated by surgery mainly through laparoscopy or methotrexate. Early and accurate diagnosis of unilateral tubal twin pregnancy prevented tubal rupture, substantial haemorrhage, and the need for emergency care, allowing elective treatment. When making a diagnosis of ectopic pregnancy, even though rare, the chance of tubal twin pregnancy has to be considered. Our case highlights the usage of transvaginal ultrasound combined with serum β -hCG in early and accurate diagnosis of ectopic pregnancy in order to prevent the risk of rupture.

References

- Gualandi M, Steemers N, Keyser JL de. [First reported case of preoperative ultrasonic diagnosis and laparoscopic treatment of unilateral, twin tubal pregnancy]. *Rev Fr Gynecol Obstet.* 1994; 89: 134-6.
- Goswami D, Agrawal N, Arora V. Twin tubal pregnancy: A large unruptured ectopic pregnancy. *J Obstet Gynaecol Res.* 2015; 41: 1820-22.
- Betti M, Vergani P, Damiani GR, Antonio P, Edoardo DN, Giuseppe T, et al. Unilateral twin tubal pregnancy: a case report and review of the literature. *Acta Biomed.* 2018; 89: 423-7.
- Saurel-Cubizolles MJ, Job-Spira N, Estryn-Behar M. Ectopic pregnancy and occupational exposure to antineoplastic drugs. *Lancet.* 1993; 341: 1169-71.
- Shaw JLV, Dey SK, Critchley HO, Horne AW. Current knowledge of the aetiology of human tubal ectopic pregnancy. *Hum Reprod Update.* 2010; 16: 432-44.
- Abusheikha N, Salha O, Brinsden P. Extra-uterine pregnancy following assisted conception treatment. *Hum Reprod Update.* 2000; 6: 80-92.
- Hanchate V, Garg A, Sheth R, Rao J, Jadhav PJ, Karayil D. Transvaginal sonographic diagnosis of live monochorionic twin ectopic pregnancy. *J Clin Ultrasound.* 2002; 30: 52-6.
- Bouyer J, Coste J, Shojaei T, Pouly JL, Gerbaud L, Job-Spira N. Risk factors for ectopic pregnancy: a comprehensive analysis based on a large case-control, population-based study in France. *Am J Epidemiol.* 2003; 157: 185-94.
- Rolle CJ, Wai CY, Bawdon R, Santos-Ramos R and Hoffman B. Unilateral twin ectopic pregnancy in a patient with a history of multiple sexually transmitted infections. *Infect Dis Obstet Gynecol.* 2006; 10306.
- Storch MP, Petrie RH. Unilateral tubal twin gestation. *Am J Obstet Gynecol.* 1976; 125: 1148.
- Goker EN, Tavmergen E, Ozcakil HT, Levi R, Adakan S. Unilateral ectopic twin pregnancy following an IVF cycle. *J Obstet Gynaecol Res.* 2001; 27: 213-5.
- Sur SD, Reddy K: Spontaneous unilateral tubal twin pregnancy. *Journal of the Royal Society of Medicine.* 2005; 98: 276.
- Urnsak I, Kadayifci O, Atay Y, Demir S, Guzel AB. A unilateral tubal twin pregnancy. *Ann Saudi Med.* 2006; 26: 333.
- Karadeniz RS, Dilbaz S, Ozkan SD. Unilateral twin tubal pregnancy successfully treated with methotrexate. *Int J Gynaecol Obstet.* 2008; 102: 171.
- Tam T, Khazaei A. Spontaneous unilateral dizygotic twin tubal pregnancy. *J Clin Ultrasound.* 2009; 37: 104-6.
- Summa B, Meinhold-Heerlein I, Bauerschlag DO, Jonat W, Mettler L, Schollmeyer T. Early detection of a twin tubal pregnancy by Doppler sonography allows fertility-conserving laparoscopic surgery. *Arch Gynecol Obstet.* 2009; 279: 87-90.
- Svirsky R, Maymon R, Vaknin Z, Mendlovic S, Weissman A, Halperin R, et al. Twin tubal pregnancy: a rising complication? *Fertil Steril.* 2010; 94: 1913-16.
- Arikan DC, Kiran G, Coskun A, Kostu B. Unilateral tubal twin ectopic pregnancy treated with single-dose methotrexate. *Arch Gynecol Obstet.* 2011; 283: 397-9.
- Fambrini M, Franchi C, Pieralli A, Sisti G, Boeri C. Spontaneous

- unilateral twin tubal pregnancy: the result of bilateral ovulation? *J Obstet Gynaecol Can.* 2012; 34: 895.
20. Samardzic D, Kasales CJ, Patrone SV. Live monochorionic adnexal twin ectopic pregnancy. *Ultrasound Q.* 2014; 30: 230-2.
 21. 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: 169-71.
 22. Ghanbarzadeh N, Nadjafi-Semnani M, Nadjafi-Semnani A, Nadj-fai-Semnani F, Shahabinejad S. Unilateral twin tubal ectopic pregnancy in a patient following tubal surgery. *J Res Med Sci.* 2015; 20: 196-8.
 23. Kim CI, Lee TY, Park ST, Kim HB, Park SH. A rare case of spontaneous live unilateral twin tubal pregnancy with both fetuses presenting with heart activities and a literature review. *Obstet Gynecol Sci.* 2018; 61: 274-7.
 24. Nepal P, Ojili V, Lu SC, Nagar A. Utility of MRI in Management of a Rare Case of Unilateral Dichorionic-Diamniotic Twin Tubal Ectopic Pregnancy. *Curr Probl Diagn Radiol.* 2019; 30142-2.
 25. Tsakiridis I, Chatzikalogiannis I, Dagklis T, Athanasiadis A, Mampopoulos A. Spontaneous unilateral twin tubal pregnancy: a rare case report with conservative management. *J Obstet Gynaecol.* 2019; 39: 1030-1.
 26. Seak CJ, Goh ZNL, Wong AC, Seak JC, Seak CK. Unilateral live twin tubal ectopic pregnancy presenting at 12 weeks of gestation: A case report. *Medicine (Baltimore).* 2019; 98: 17229.
 27. Lategan HE, Gillispie VC. Spontaneous Unilateral Tubal Twin Ectopic Pregnancy. *Ochsner J.* 2019; 19: 178-80.