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

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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 preoperatively [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 dizygotic 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

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

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

<table>
<thead>
<tr>
<th>Author</th>
<th>Clinical symptoms</th>
<th>Risk factors</th>
<th>Treatments</th>
<th>Monozygotic/ dizygotic</th>
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</thead>
<tbody>
<tr>
<td>Goker et al(11)</td>
<td>Vaginal bleeding</td>
<td>Infertility (IVF)</td>
<td>Lap salpingectomy</td>
<td>NR</td>
</tr>
<tr>
<td>Hanchate et al(7)</td>
<td>Vaginal bleeding and pain</td>
<td>No</td>
<td>Paro salpingectomy</td>
<td>Monochorionic</td>
</tr>
<tr>
<td>Sur et al(12)</td>
<td>Pain and vaginal bleeding</td>
<td>Chlamydia infection</td>
<td>Lap salpingectomy</td>
<td>Dichorionic</td>
</tr>
<tr>
<td>Urunsk et al(13)</td>
<td>Pain and vaginal bleeding</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Karadeniz et al(14)</td>
<td>Vaginal bleeding and pain</td>
<td>Infertility (ART)</td>
<td>MTX</td>
<td>NR</td>
</tr>
<tr>
<td>Tam et(15)</td>
<td>Pain</td>
<td>Tubal surgery</td>
<td>Paro salpingectomy</td>
<td>Dichorionic</td>
</tr>
<tr>
<td>Summa et al(16)</td>
<td>No</td>
<td>No</td>
<td>Lap tube anastomosis</td>
<td>Monochorionic</td>
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<tr>
<td>Svirsky et al(17)</td>
<td>Suspicion of tubal pregnancy</td>
<td>No</td>
<td>Infertility (IVF)</td>
<td>Lap salpingectomy</td>
</tr>
<tr>
<td>Arikan et al(18)</td>
<td>Vaginal bleeding and pain</td>
<td>Infertility</td>
<td>MTX</td>
<td>Dichorionic</td>
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<td>Fambrini et al(19)</td>
<td>Pain and vaginal bleeding</td>
<td>No</td>
<td>Lap salpingectomy</td>
<td>Dichorionic</td>
</tr>
<tr>
<td>Samardzic et al(20)</td>
<td>Pain and nausea</td>
<td>Smoking</td>
<td>Lap salpingectomy</td>
<td>Monochorionic</td>
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<td>Longoria et al(21)</td>
<td>No</td>
<td>Tubal surgery</td>
<td>Lap salpingectomy</td>
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<td>Ghanbarzadeh et al(22)</td>
<td>Pain and vaginal bleeding</td>
<td>Tubal surgery</td>
<td>Paro salpingectomy</td>
<td>Monochorionic</td>
</tr>
<tr>
<td>Kim et al(23)</td>
<td>Nausea, vomiting, and pain</td>
<td>Abortion</td>
<td>Right salpingectomy</td>
<td>Dichorionic</td>
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<td>Betti et al(3)</td>
<td>No</td>
<td>Tubal surgery</td>
<td>MTX, Lap salpingectomy</td>
<td>NR</td>
</tr>
<tr>
<td>Nepal et al(24)</td>
<td>Abdominal pain</td>
<td>Infertility (IVF)</td>
<td>Lap salpingoophorectomy</td>
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<tr>
<td>Tsakiridis et al(25)</td>
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<td>No</td>
<td>MTX</td>
<td>NR</td>
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<td>Seak et al(26)</td>
<td>Pain</td>
<td>No</td>
<td>Lap salpingectomy</td>
<td>Monochorionic</td>
</tr>
<tr>
<td>Lategan et al(27)</td>
<td>Vaginal bleeding and pain</td>
<td>Infertility (IVF)</td>
<td>Lap salpingostomy</td>
<td>NR</td>
</tr>
<tr>
<td>Present study</td>
<td>No</td>
<td>Abortion</td>
<td>Lap salpingectomy</td>
<td>Dichorionic</td>
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</table>
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 pregnancy. More than half cases (57.89%) were nulliparous. 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


