

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

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Abstract:

Unilateral twin tubal pregnancy is rare with an incidence of 1:1,25,000 spontaneous pregnancies. Majority of cases occur following ovulation induction and assisted reproductive technology. Twin tubal ectopic pregnancies are rare forms of ectopic pregnancies, with an incidence of one in 200 ectopic pregnancies. We present a rare case of ruptured ectopic pregnancy which occurred due to unilateral twin tubal pregnancy. This was a spontaneous pregnancy with no risk factor for ectopic pregnancy. An emergency laparotomy with right sided salpingectomy was performed, with an uncomplicated post operative period. The diagnosis was arrived at via trans-abdominal pelvic ultrasonography and confirmed postoperatively with histology. A high index of suspicion is required in the diagnosis of ectopic gestation in order to reduce the morbidity associated with it. Unilateral twin tubal ectopic gestation, although rare, has been reported with higher frequencies, and as such, the occurrence should be carefully sought on ultrasound in order to reduce its potential morbidity and mortality.

Keywords: Ectopic pregnancy, spontaneous unilateral twin tubal ectopic pregnancy, laparotomy, salpingectomy.

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I. Introduction:

An ectopic gestation occurs when a blastocyst gets implanted in a location other than the endometrial lining of the uterus. The incidence of ectopic gestations has increased since the advent of assisted reproductive techniques (ART) and tubal surgeries(1). It occurs in 1–2% of first trimester pregnancies in the general population but increases to 2–5% in patients who have utilized ART(2,3). The most frequent type of ectopic twin pregnancy is heterotopic pregnancy that has increased following *in vitro* fertilization(4). Spontaneous unilateral twin tubal ectopic pregnancy is a very rare occurrence. It is estimated to have an incidence of 1:20,000–1:1,25,000 spontaneous pregnancies and 1:200 ectopic pregnancies(5,6). This is a case of ruptured unilateral twin tubal ectopic gestation which was diagnosed prior to emergency surgery via transabdominal pelvic ultrasonography and confirmed with histology.

II. Case Report :

A 25-years-old lady P1+1 ,G3 presented to our emergency in the Dept. of Obstetrics and Gynecology of Bankura Sammilani Medical College and Hospital, Bankura, West Bengal, India with the chief complaint of lower abdominal pain followed by bleeding per vagina associated with nausea and vomiting for 3 days with a history of 2 months amenorrhea prior to presentation.

On admission, she was alert, anxious, cooperative and in moderate shock. Her blood pressure (BP) was 100/70 mmHg, with high pulse rate 110/min, temperature normal, with moderate amount of pallor.

She had a history of one caesarean section 7 years back with a living child and subsequently one spontaneous abortion. The patient had received no infertility treatments and had no risk factors for ectopic pregnancy. Appendectomy was done 4 years back.

On per abdominal examination, abdomen was tensed with muscle guarding with tenderness in right iliac fossa. Per vaginal examination showed bulky uterus with parous os, tenderness in the right fornix with an

adnexal mass. Cervical motion tenderness was seen positive. Finger was blood stained during per vaginal examination .

Urine for pregnancy test was positive. Before operation hemoglobin level was 6 gm%. The other blood chemistry levels were examined later. Urgent ultrasonography (TAS+TVS) report revealed right-sided tuboovarian mass adjacent to the uterus and right adnexa with a single gestational sac containing two fetal poles of 9 weeks each with ?? cardiac activity. Echogenic free fluid in pelvis, abdomen extending up to hepatorenal pouch, splenorenal recess. Endometrial cavity was empty- suggestive of disturbed tubal ectopic pregnancy with two fetal poles (**Figure 1**).



Figure 1: Ultrasound revealed an empty uterus and 2 fetuses in a single gestational sac in the right adnexa.

A diagnosis of ruptured twin tubal ectopic pregnancy was made. The patient was prepared for urgent exploratory laparotomy and sent to operation theatre immediately. Simultaneously resuscitation was going on to correct the shock. Under general anesthesia abdomen was opened. After opening abdomen, hemoperitoneum (approximately 1Litre) detected with right ampullary ectopic mass of size 4×3 cm which had ruptured and actively bleed. Two fetuses along with some part of placental tissue were seen coming out from the ruptured site. Uterus was bulky. Left sided tube was intact and healthy. Both ovaries were healthy (**Figure 2 &3**).

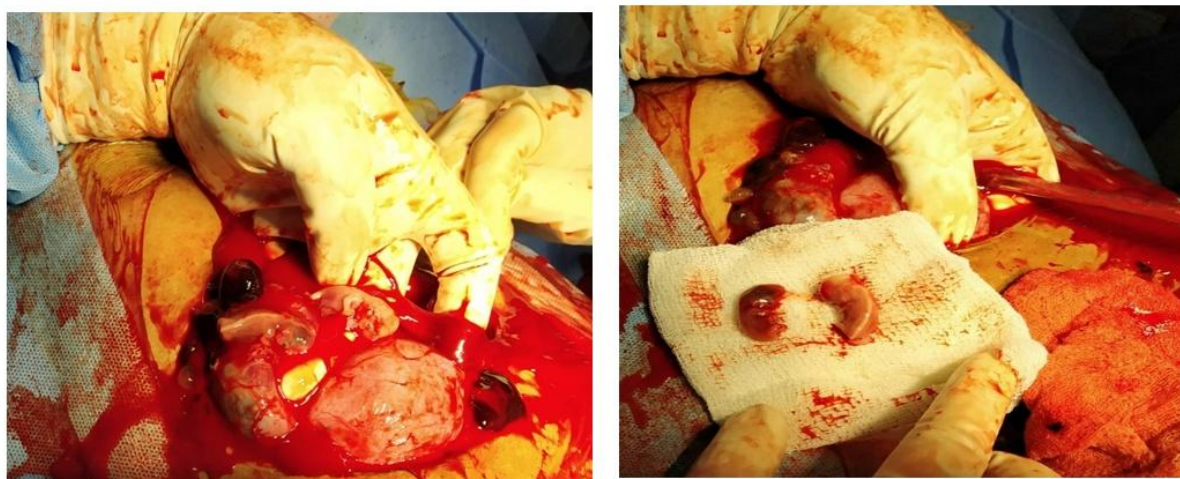


Figure 2: A 4x3 cm ectopic mass in the right adnexa with rent and bleeding from the ampullary region. Two fetuses along with some part of placental tissue were seen coming out from the ruptured site.



Figure 3: Right ampullary ectopic mass and twin fetuses.



Figure 4 : Right sided salpingectomy specimen with placenta and clot.

Right-sided salpingectomy was done and hemostasis was secured(**Figure 4**). Abdomen was closed in layers after peritoneal wash, placing abdominal drain and taking count of the instruments and the mops. Specimen of right-sided fallopian tube with ectopic mass along with two fetuses were sent for histopathological examination. Postoperative period was uneventful. Four units of whole blood were transfused. Patient was discharged in satisfactory condition after stitch removal. Histopathological examination of the tube showed the features of tubal gestation.

III. Discussion

Although twin tubal pregnancy was thought to be very rare, its incidence has increased. Assisted reproductive technology (ART) is responsible for the increase in twin tubal pregnancy. Unilateral twin tubal gestations are extremely rare with a reported incidence of one per 200 ectopic pregnancies or one per 1,25,000 spontaneous pregnancies(**5,6**). It was first described in 1891 by De Ott(**7**). Spontaneous live unilateral twin tubal pregnancy is extremely rare in comparison with bilateral twin tubal pregnancy and unilateral twin tubal pregnancy with no cardiac activity(**8,9**). In 1994 , Gualandi et al. documented the first case of unilateral, tubal twin pregnancy with cardiac activity in both embryos, by endovaginal ultrasound(**10**). There were less than 12 unilateral ectopic twin pregnancies reported with beating hearts in both embryos(**5**). In our case the sonologist was uncertain about the viability of the fetuses.

Many risk factors exist for ectopic pregnancies and include genital tract infections, tubal surgeries, previous ectopic gestations (with history of recurrence of 12–18%), use of fertility drugs and ART, increasing

maternal age, smoking, and congenital uterine anomalies, intrauterine device(1). Most of the cases occur following ART (5,6) with very few cases occurring spontaneously as in our case.

Anything that interferes with ovum transport in the fallopian tube increases the risk for ectopic gestation. Also, a delay in ovum transport and implantation in monozygotic twinning may increase the risk for ectopic gestation. The size of the twin cell mass has been hypothesized to cause retardation in transport. The zygosity of twin in the present case was not studied. Most of the twin tubal pregnancies were thought to be monozygotic. However, in the majority of the reported cases, the zygosity was determined subjectively by using observation such as size and gestational age similarity, fetal membranes and number of corpus luteum (11). It seems to be that the method to determine zygosity of the twins by using DNA probes that detect restriction fragment length polymorphisms more accurately (12). In this case the gestational age of both fetuses was same i.e 9 weeks.

As such, about 95% of twin ectopic gestations reported have been monochorionic and monoamniotic(6). This index case was monoamniotic.

Monochorionic, monoamniotic twin pregnancies will be unilateral. However, if it is dichorionic, diamniotic, it may be unilateral but may rarely present as a bilateral ectopic(13). In our case the ectopic was unilateral.

The advent of transvaginal ultrasonography and correlation with quantitative serum β -hCG evaluation has improved the detection rates of ectopic gestations (1,5). The unstable hemodynamic status of the patient in this index case did not allow time for quantitative evaluation of serum β -hCG, hence, a spot pregnancy test was done. The diagnosis was also arrived at via trans-abdominal pelvic ultrasonography and confirmed postoperatively with histology.

Despite declining rates of morbidity and mortality following ectopic gestations due to improved diagnosis and treatment modalities, it still remains a potentially life threatening condition. The risk of complication is higher in twin tubal pregnancies compared with single tubal pregnancies because 2 gestational sacs with 2 fetuses would cause greater volume expansion of the tubal implantation site than 1 gestational sac with 1 fetus. The risk of acute abdomen and hypovolemic shock due to rupture of one or both of the tubes is 30% to 50% in twin tubal pregnancies (5). Therefore, in twin tubal pregnancy, early recognition, accurate diagnosis, and immediate treatment are very important to minimize the morbidity and mortality. When the diagnosis of ectopic pregnancy is established, especially when the patient conceived with ART, the possibility of developing multiple ectopic pregnancies should be considered, and effort should be exerted to find the probable ectopic foci (8).

Treatment of an ectopic pregnancy depends on its clinical presentation, size of the ectopic mass and β -hCG levels. Surgical management is done in acute ruptured ectopic pregnancy, in haemodynamically unstable patient or in those who have failed medical treatment or have contraindications to medical treatment. Laparoscopic is the preferred treatment as it is associated with lower cost, less operating time, shorter hospital stays and faster recovery. Salpingectomy is the recommended treatment; however, salpingostomy can be considered for women with one tube who are wishing to preserve their fertility.

This patient had an acute abdomen from tubal rupture. Treatment was done with laparotomy and right sided salpingectomy as opposed to the laparoscopic approach employed in most of the recent literature. This was done due to the unavailability of facilities for emergency laparoscopic surgery in our hospital. We did salpingectomy as the rent was big.

This patient conceived spontaneously without ART, whereas most cases of twin tubal pregnancies were associated with ART and other risk factors. This case is reported for its rarity and also signifies the need of early ultrasound (preferably transvaginal ultrasound) for diagnosing ectopic pregnancy even in low risk women.

IV. Conclusion :

The incidence of unilateral twin tubal pregnancy may not be as low as earlier perceived. Early diagnosis and prompt management would forestall the morbidity associated with this condition and as such meticulous and detailed ultrasonography (preferably transvaginal ultrasound) should be employed early in the first trimester to aid diagnosis. This could lead to the use of other less invasive forms of treatment as well as reducing morbidity associated with it.

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Conflicts of interest :

There are no conflicts of interest.

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