DOI: 10.7860/JCDR/2016/16521.7416

Case Report

Obstetrics and Gynaecology Section

Bilateral Simultaneous Tubal Ectopic Pregnancy: A Case Report, Review of Literature and a Proposed Management Algorithm

SAUBHAGYA KUMAR JENA[1], SWETA SINGH[2], MONALISHA NAYAK[3], LEENA DAS[4], SWAGATIKA SENAPATI[5]

ABSTRACT

Bilateral simultaneous Tubal Ectopic Pregnancy (BTP) is the rarest form of ectopic pregnancy. The incidence is higher in women undergoing assisted reproductive techniques or ovulation induction. The clinical presentation is unpredictable and there are no unique features to distinguish it from unilateral ectopic pregnancy. BTP continues to be a clinician's dilemma as pre-operative diagnosis is difficult and is commonly made during surgery. Treatment options are varied depending on site of ectopic pregnancy, extent of tubal damage and requirement of future fertility. We report a case of BTP which was diagnosed during surgery and propose an algorithm for management of such patients.

Keywords: Bilateral ectopic pregnancy, β-hCG, Salpingectomy, salpingostomy

CASE REPORT

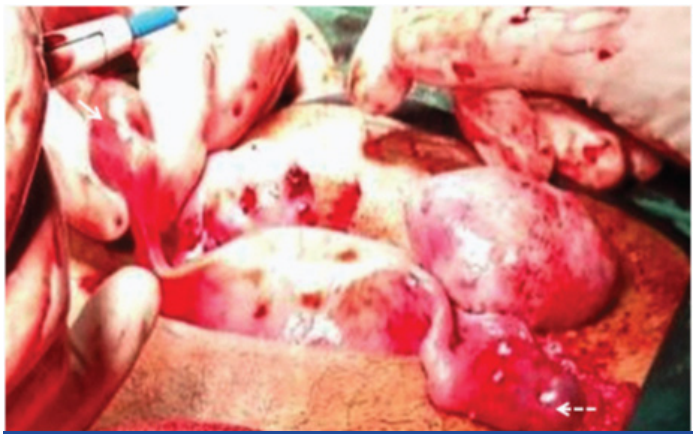
A 23-year-old, para one, unbooked case, presented to our institute with history of amenorrhoea of 7 weeks and 5 days and spotting per vaginum for the past 9 days. She had consulted a local private practitioner the previous day and urine pregnancy test had been done, which was positive. She had a full term normal delivery 3 years back, which was a spontaneous conception. On history it was revealed that the patient was trying for natural conception for the past 3 months. Her previous menstrual cycles were regular and there was no history of pelvic inflammatory disease (PID), tuberculosis, previous abdominopelvic surgery or use of any contraception. She fallaciously denied any history of having received infertility treatment at this moment. Patient had an abdominal ultrasonography report done from outside the previous day, which was suggestive of chronic left-sided tubal ectopic pregnancy.

On examination, there was moderate degree of pallor with tachycardia. Her blood pressure measured 90/60 mmHg with a regular heart rate of 115-120 beats/minute. There was diffuse abdominal tenderness but no guarding or rigidity. Mild bleeding was present through the os. Per vaginum examination revealed a normal sized uterus, with fullness in the Pouch of Douglas (POD) and a 4 x 3 cm tender mass in the right fornix. A repeat urine pregnancy test was positive at our institute. Emergency abdominal ultrasonography was done at our institute, which revealed an empty uterus with a heterogeneous mass of size 6.6 x 4.8 cm in the POD and moderate amount of collection in the pelvis suggestive of blood. A provisional diagnosis of right sided ruptured tubal ectopic pregnancy was made as evidenced on current ultrasound and emergency laparotomy performed.

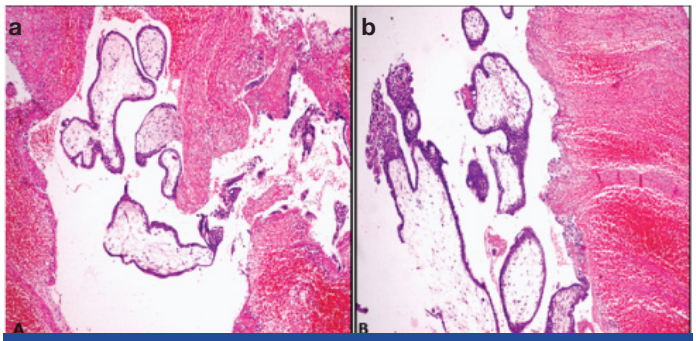
At laparotomy, there was 100 ml of haemoperitoneum with approximately 500 ml of clots in the POD. The right tube was the seat of a 2 x 3 cm tubal ectopic pregnancy in the ampullary region, with active ongoing bleeding from the fimbrial end suggestive of tubal abortion [Table/Fig-1]. The uterus was normal in size. On inspection of the left tube, there was another mass of 3 x 3 cm in the ampullary region with a thinned out bulging point suggestive of near rupture [Table/Fig-1]. Both the ovaries were enlarged and had a solid appearance, measuring approximately 4 x 3 cm, raising suspicion of possible ovulation induction. An option of salpingectomy on right side and salpingostomy on left side was discussed with the relatives, but they wished for conservative management of both tubes for preserving future fertility. The possibility of future recurrent ectopic pregnancy with conservative surgery was explained to them.

Bilateral salpingostomy was performed and the expressed products of conception from both tubes were sent for histo-pathological examination, which later confirmed the presence of chorionic villi in both tubes [Table/Fig-2a&b]. Her postoperative period was uneventful, and she was discharged on day 10 in stable condition with an advice for serial β-human chorionic gonadotrophin (β-hCG) monitoring to rule out a persistent trophoblastic disease. Before discharge, on questioning the patient, she revealed the treatment papers from the previous practitioner which showed that she had received tablet clomiphene citrate 100 mg from days 5 to 9 and injection follicle stimulating hormone on alternate days from days 5 to 9; intrauterine insemination had been done on day 14 during the cycle that she conceived.

Her baseline β-hCG was 9602 IU/L which decreased to 172 IU/L on day 7 and to 11.2 IU/L on day 13. One month after discharge, her β-hCG was < 5IU/L. Follow up abdominal ultrasound examination revealed a normal sized uterus and a decrease in the size of ovary. Hysterosalpingography done 6 months later showed both tubes to



[Table/Fig-1]: Uterus with right fallopian tube showing ampullary ectopic pregnancy and active bleeding from its fimbrial end (right sided tubal abortion) (arrow) and an unruptured left tubal pregnancy (dashed arrow).



[Table/Fig-2]: Histopathology of POC: a) from right tube; and b) from left tube showing chorionic villi diagnostic of bilateral tubal ectopic pregnancy; POC- products of conception

be patent with bilateral free spill. Presently, the patient is 9 months postoperative and has not yet conceived.

DISCUSSION

While unilateral tubal ectopic pregnancy is the commonest form of ectopic gestation, simultaneous Bilateral Tubal ectopic Pregnancy (BTP) is the rarest variety, with a reported incidence of 5 in 1 million deliveries [1]. Higher incidence of BTPs has been seen after the use of Assisted Reproductive Techniques (ARTs) or following ovulation induction [2].

In the absence of ARTs or ovulation induction, BTP is the rarest form of extra uterine pregnancy [1]; twin pregnancies in the same tube and heterotopic pregnancies are thought to be more common [3,4]. The incidence of BTP is 1 in 725 to 1580 ectopic pregnancies [5], corresponding to 1 per every 200,000 live births [6]. It is possible that the actual incidence of BTPs may be higher but many cases go unrecognised or unreported. Furthermore, risk factors for the occurrence of ectopic gestation like PID, sexually transmitted diseases, use of antibiotics for treating PID instead of salpingectomy, ovulation induction and tubal surgeries are quite common [1].

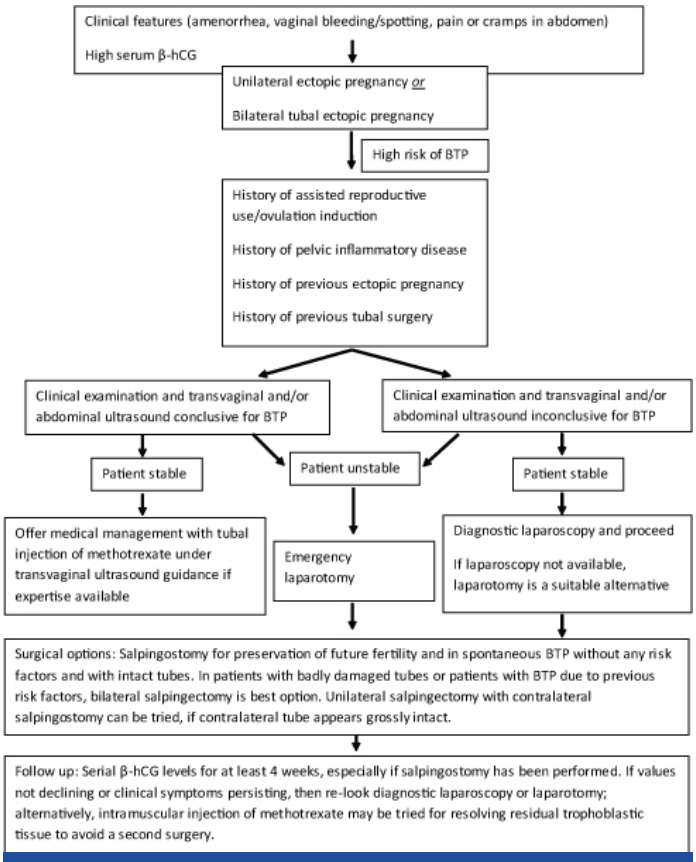
The mechanisms of BTP have been postulated variously as multiple ovulations, transperitoneal migration of trophoblastic tissue from one tube to the other and superfetation [7,8]. The diagnostic criteria were first laid out by Fishback who suggested that there should be a description of foetal parts or foetus as well as placental parts from both the tubes [5]. This was later modified by Norris, who stated that microscopic demonstration of chorionic villi in both tubes was sufficient for the diagnosis [9]. Thus, in most cases, the diagnosis remains essentially surgical or postoperative [9].

Neither clinical symptoms (triad of amenorrhea, vaginal bleeding and pain abdomen) nor serum β-hCG estimation can reliably differentiate BTP from unilateral tubal ectopic pregnancy. Ultrasonography has only rarely picked up BTP preoperatively and that too, in the presence of live embryos in both tubes [10]. Presence of unilateral ectopic pregnancy or adnexal masses has the same clinical presentation as BTP and therefore, proper interrogation of the other tube with ultrasound may be commonly missed [10]. Thus, ultrasound cannot be advocated as standard of care in the diagnosis of this condition [1].

The management varies depending upon the condition of the patient, extent of tubal damage and the wish for future fertility [1]. Medical management with methotrexate successfully injected consecutively into each tube under transvaginal ultrasound guidance has been described, where the diagnosis of BTP was made preoperatively [10]. However, it is essential to diagnose BTP accurately, as failure of medical management with administration of single dose inj. methotrexate for presumed unilateral tubal ectopic pregnancy has been reported in a case which later turned out to be BTP [11]. Intramuscular injection of methotrexate in a dose of 50 mg/m^2 has also been used to resolve persistent ectopic pregnancy after laparoscopic bilateral salpingostomy for BTP [10].

Surgical management has ranged from salpingectomy for one tube and linear salpingostomy for the other, to bilateral salpingostomy or bilateral salpingectomy [6]. If available, laparoscopy may be the best option both for diagnosis and management of BTP [12]. However, one needs to keep a high index of suspicion for BTP as the diagnosis can be easily missed even on laparoscopy [12]. Laparotomy is equally effective, and is the management of choice if patient is haemodynamically unstable. Of note however, serial β-hCG monitoring should be done as there is a high chance of persistent ectopic pregnancy, especially if a conservative surgery like salpingostomy or milking of the tube has been performed [1]. Reports of intrauterine pregnancy exist after conservative surgical management of BTP [6,7], but the fact remains that these patients are at high risk for recurrent ectopic pregnancies in the future [1].

In the absence of any treatment guidelines, we propose a simple management algorithm for women presenting with possible BTP [Table/Fig-3], so as to allow a methodical approach in order to minimize the possibility of missing or misdiagnosing BTP.



[Table/Fig-3]: Algorithm for management of suspected Bilateral Tubal ectopic Pregnancy (BTP)

CONCLUSION

BTP represents a clinician's conundrum as the presentation is quite similar to that of unilateral ectopic pregnancy and is easily missed. Furthermore, there are no treatment guidelines or protocols available for the management of this rare clinical entity. Young parous women who have been trying for conception less than 12 months should not be offered ovulation induction and intra-uterine insemination, as it may lead to BTP as a result of ovarian hyper-stimulation, as illustrated by this case. To conclude, a high index of suspicion and thorough inspection of both tubes even in the presence of dense adhesions, either during diagnostic laparoscopy and/or definitive surgery are simple measures to avoid missing this rare life threatening condition.

REFERENCES

[1] Andrews J, Farrell S. Spontaneous bilateral tubal pregnancies: Acase report. *J Obstet Gynaecol Can*. 2008;30:51-54.
[2] Brady J, Wilson M. Spontaneous bilateral tubal ectopic pregnancy. *J R Soc Med*. 2005;98:120–21.
[3] Norris S. Bilateral simultaneous tubal pregnancy. *Can Med Assoc J*. 1953;68:379–81.
[4] Fox EJ, Mevs FF. Simultaneous bilateral tubal pregnancies. Report of 2 cases. *Obstet Gynecol*. 1963;21:499–501.
[5] Fishback HR. Bilateral simultaneous tubal pregnancy. *Am J Obstet Gynecol*. 1939;37:1035.
[6] Edelstein MC, Morgan MA. Bilateral simultaneous tubal pregnancy: case report and review of the literature. *Obstet Gynecol Surv.* 1989;44:250–52.
[7] Foster HM, Lakshin AS, Taylor WF. Bilateral tubal pregnancy with vaginal delivery. *Obstet Gynecol*. 1982;60:664–66.
[8] Tabachnikoff RM, Dada MO, Woods RJ, Rohere D, Myers CP. Bilateral tubal pregnancy. A report of an unusual case. *J Reprod Med.* 1998;43:707–09.
[9] Norris S. Bilateral simultaneous tubal pregnancy. *Can Med Assoc J.* 1953;68:379–81.
[10] Sentilhes L, Bouet PE, Jalle T, Boussion F, Lefebvre-Lacoeuille C, Descamps P. Ultrasound diagnosis of spontaneous bilateral tubal pregnancy. *Aust NZJ Obstet Gynaecol*. 2009;49:695-96.
[11] Marcovici I, Scoccia B. Spontaneous bilateral tubal ectopic pregnancy and failed methotrexate therapy: a case report. *Am J Obstet Gynecol*. 1997;177:1545-46.
[12] Li W, Wang G, Lin T, Sun W. Misdiagnosis of bilateral tubal pregnancy: a case report. *J Med Case Rep*. 2014;8:342.

PARTICULARS OF CONTRIBUTORS:
1. Assistant Professor, Department of Obstetrics and Gynaecology, All India Institute of Medical Sciences (AIIMS), Bhubaneswar, Odisha, India.
2. Assistant Professor, Department of Obstetrics and Gynaecology, All India Institute of Medical Sciences (AIIMS), Bhubaneswar, Odisha, India.
3. Senior Resident, Department of Obstetrics and Gynaecology, All India Institute of Medical Sciences (AIIMS), Bhubaneswar, Odisha, India.
4. Senior Resident, Department of Obstetrics and Gynaecology, All India Institute of Medical Sciences (AIIMS), Bhubaneswar, Odisha, India.
5. Senior Resident, Department of Pathology, All India Institute of Medical Sciences (AIIMS), Bhubaneswar, Odisha, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Sweta Singh,
Assistant Professor, Department of Obstetrics and Gynaecology, AIIMS Bhubaneswar-751019, Odisha, India.
E-mail: swetsingh@hotmail.com

FINANCIAL OR OTHER COMPETING INTERESTS: None.

Date of Submission: **Aug 28, 2015**
Date of Peer Review: **Nov 01, 2015**
Date of Acceptance: **Dec 16, 2015**
Date of Publishing: **Mar 01, 2016**