

Spontaneous bilateral tubal pregnancy in a nulliparous woman. Laparoscopic diagnosis and treatment

Gloria Calagna¹, Claudio Rossi², Giorgio Adile¹, Maria Manzone¹, Antonino Perino¹, Gaspare Cucinella¹.

¹Department of Obstetrics and Gynaecology, University Hospital "P. Giaccone", Palermo (Italy)

²Obstetrics and Gynecology, "Villa Sofia-Cervello" Hospital, Palermo (Italy).

ABSTRACT

A 34 year-old nulliparous woman was referred to our Department, complaining of vaginal bleeding and intermittent lower abdominal cramping. serum level of β -hCG was 4954 IU/L. Transvaginal ultrasound examination revealed no gestational sac in the uterine cavity and a right adnexal mass, suggestive of ectopic pregnancy; left adnexal area appeared regular. At laparoscopy, the presence of an ectopic pregnancy was confirmed in the right tube; on the left side, surprisingly, there was an intact infundibular ectopic pregnancy. Right tuba was removed; considering the condition of nulliparous of the patient, we decided to perform a linear salpingostomy. Histologic exam confirmed the diagnosis of spontaneous bilateral tubal ectopic pregnancies.

Keywords: Bilateral tubal pregnancy, ectopic pregnancy, laparoscopy, salpingostomy

SOMMARIO

Una donna di 34 anni, nullipara, giunse alla nostra osservazione, lamentando sanguinamento vaginale e crampi addominali intermittenti. Il dosaggio della β -hCG risultava 4954 IU/L. L'esame ecografico transvaginale rivelava assenza di sacco gestazionale in cavità uterina e l'immagine di una massa annessiale destra, suggestiva di gravidanza ectopica. La laparoscopia confermava la presenza di un gravidanza ectopica nella tuba di destra; nel lato sinistro, sorprendentemente, si evidenziava una seconda gravidanza ectopica infundibolare, intatta. E' stata effettuata una salpingectomia destra; in considerazione della condizione di nulliparità della paziente, abbiamo deciso di effettuare una salpingostomia lineare. L'esame istologico ha confermato la diagnosi di gravidanza tubarica bilaterale spontanea.

INTRODUCTION

In the absence of preceding induction of ovulation, bilateral tubal pregnancy (BTP) is an extremely unusual occurrence. Its frequency has been estimated at 1/200 000 uterine pregnancies and 1/725–1/1580 ectopic pregnancies⁽¹⁾. However, in the last decades a 3-fold increase in the incidence of BTP as well as heterotopic pregnancy has been observed, mainly related to the rise of assisted reproduction techniques (ARTs)^(2,3). Preoperative diagnosis of BTP remains a challenge. Serum β -hCG estimation is not reliable as the values will be elevated more than that of a single ectopic. Detection with ultrasound scan is almost difficult and commonly failed in achieving a correct preoperative diagnosis⁽⁴⁾. We report a misdiagnosed case of spontaneous BTP managed laparoscopically

CASE REPORT

A 34 year-old nulliparous woman was referred to our gynaecology Unit with an approximate gestational age of 8 weeks, complaining of vaginal bleeding and intermittent lower abdominal cramping. She had no history of oral contraceptives, intrauterine device (IUD) or fertility drugs use, nor pelvic inflammatory disease and abdominal/pelvic surgery. Her menstrual history was not relevant. Interestingly, both patient and her husband had a family history of twins.

A physical examination revealed stable vital signs (blood pressure: 100/60 mmHg; heart rate: 75 beats/minute). Abdomen was soft and non-tender on palpation. At bimanual vaginal examination, we noted a bulky uterus and the adnexa were difficult to palpate; moreover, vaginal bleeding was confirmed. Admission serum level of β -hCG was 4954IU/L. Transvaginal ultrasound (US) examination revealed no gestational sac in the uterine cavity and a right inhomogeneous adnexal

Correspondence to: gloria.calagna83@gmail.com

Copyright 2015, Partner-Graf srl, Prato

DOI: 10.14660/2385-0868-27

mass, suggestive of ectopic pregnancy; nothing abnormal was detected in the left adnexal area. A small amount of fluid was present in the pouch of Douglas. The presumptive diagnosis of right tubal pregnancy was made.

After counseling with the patient with detailed explanation of treatment options, a laparoscopy was performed. Intra-operatively, a small amount of bleeding was noted in the anterior and posterior cul-de-sac. Uterus and ovaries were regular in morphology. The right tube showed an intact ectopic pregnancy (about 35x35 mm) in the ampullary region; on the left side, surprisingly, there was an intact infundibular ectopic pregnancy (measuring 20x20 mm approximately), that was bleeding and forming an organized haematoma between the fimbriae (**Figure 1**). Patient had spontaneous bilateral tubal pregnancies.

The right tube appeared greatly distended and, during the phase of its surgical mobilization, the rupture of the tubal wall, in its lower part, has been caused. Consequently, we decided to remove it, thinking that the the risks of conservative management were higher than the possible benefits



Figure 1.
Intra-operative view of BTP.

However, considering the condition of nulliparous of the patient, we decided to perform a linear tube salpingostomy with cautery on the left tube, in order to preserve future fertility (**Figure 2**); a small product of conception was retrieved from the incision. No intra-operative complications occurred.



Figure 2.
Detail of the intact left tubal pregnancy (U: uterus; Ov: ovary; TP: tubal pregnancy).

All the specimens were sent for histologic exam, which revealed the presence of inflamed decidua with chorionic villi in both tubes. On day 1, a single-dose methotrexate (MTX) injection (50 mg/m²) was given to reduce the risk of post-salpingostomy ectopic pregnancy persistence. We monitored β -hCG level following the single-dose scheme of MTX treatment: on the 7th day, β -hCG declined to 294.3 UI/ml and 2 weeks after surgery, it was negative. At 6 months, patient was fine and β -hCG was negative.

DISCUSSION

Ectopic pregnancies are known to occur with increased frequency after ARTs and, accordingly, the rare event of BTP is generally related to the in vitro fertilization - embryo transfer (IVF-ET) treatment, as reported in medical literature^(2,5,6). Cases of heterotopic triplet pregnancy (BTP and

intrauterine pregnancy) after IVF-ET are also described⁽³⁾.

However, in many cases of BTP, no traditional risk factors are identified and these cases are called “primitive” or “spontaneous”⁽⁷⁻⁹⁾. The real mechanism to explain spontaneous BTP events is unclear. Among the hypotheses, multiple ovulation and trans-peritoneal migration of trophoblastic tissue from one tube to another, are included^(10,11).

Pre-operative diagnosis of BTP is, to date, a daunting task. Clinical presentation is generally non-specific and unpredictable, and above all there are no specific clinical features to distinguish a BTP from a unilateral tubal form. Moreover, transvaginal US hardly ever allows clear identification of BTP; sometimes a visible tubal pregnancy that coexists with a doubtful image in the contralateral adnexa can arouse suspicion. Finally, serum levels of β -hCG and progesterone cannot distinguish BTP from a unilateral ectopic pregnancy.

Consequently, BTP is diagnosed in the operating room, when a second gestational sac on the contralateral tube is noted during direct inspection. However, a histopathological examination is essential to obtain a definitive

diagnosis. Main diagnostic criterion of BTP, first suggested by Fishblack in 1939⁽¹²⁾ and then confirmed by Norris in 1956⁽¹³⁾, stated that the histologic presence of chorionic villi in each tubes should be sufficient to justify the diagnosis.

The management of bilateral tubal ectopic pregnancies may be either surgical (salpingostomy or salpingectomy) or medical (methotrexate administration), and depends to a large extent on the clinical status of the patient and the state of the fallopian tubes at presentation. Use of methotrexate is associated with a significant risk of failure in the treatment of BTP⁽¹⁴⁾. Therefore, a surgical approach is currently preferred.

In the last few years, with the advancements and diffusion of mini-invasive surgery⁽¹⁵⁾, laparoscopy has become the gold standard for both diagnosis and treatment of this pathologic condition. The specific technical approach to choose depends on two main factors: 1. age of the patient and the desire of future fertility; 2. status of the involved tubes. Performing a review of international literature (including only articles in English language), we identified six papers describing laparoscopic management of spontaneous BTP^(4,16-20) (Table 1). All the include articles reported a single case.

Table 1.
Review of the literature on laparoscopic treatment of spontaneous BTP.

Refrence	No. of cases (Age)	Admission symtoms	Admission serum level of β -hCG	Type of surgery	Adjunctive medical therapy	Histology	Note
Idotta et al, 2001 (16)	1 (28)	no ymptoms	not reported	bilateral salpingotomy	no	chorionic villi in each tube.	previous conservative laparoscopy for tubal pregnancy and laparotomy for corpus luteal hemorrhage.
Sommer et al, 2002 (17)	1 (30)	painless vaginal bleeding	966 IU/L	bilateral salpingotomy	no	chorionic villi in each tube.	recurrent left-side tubal pregnancy (12 months after).
Andrews et al, 2008 (18)	1 (25)	vaginal bleeding, lower abdominal cramping	24.242 IU/L	bilateral salpingostomy	yes (methotrexate 50 mg/m ²)	chorionic villi in each tube.	fragments of fetal tissue (vertebral column, neurological structures, liver, intestine, umbilical cord) from LT.
Liao et al, 2009 (19)	1 (30)	vaginal spotting, pelvic pain	686 IU/L	bilateral salpingostomy	no	not reported	lysis of adhesions and left fimbrioplasty was also performed.
Mathlouthi et al, 2012 (20)	1 (37)	vaginal bleeding, pelvic pain signs of collapse	15.73 mIU/mL	salpingectomy (LT); salpingostomy (RT).	no	immature placental villi (RT); placental tissue (LT).	LT was ruptured; RT was unruptured.
Li et al, 2014 (22)	1 (33)	vaginal bleeding	6993.1 IU/L	isthmus resection (RT); salpingostomy (LT).	no	placental villi in each tube.	left tubal pregnancy eight years before treated with methotrexate

Note.
LT: left tube; RT: right tube.

Most patients were nulliparous (4/6, 66.7%) and, consequently, in most cases authors performed a conservative surgical approach (5/6, 83.3%). In only one case, a recurrent tubal pregnancy occurred (12 months after conservative treatment)⁽¹⁷⁾.

In the case of substantial damage or active bleeding of one or both tubes, salpingectomy seems to be the most proper and safest treatment. In young and nulliparous women, preservation of at least one tube should be the main goal of the treatment and, as in the present reported case, salpingostomy should to be preferred. Spontaneous conception after BTP surgery is difficult; however, subsequent viable intrauterine pregnancies after salpingostomy were described⁽²¹⁾.

Currently, there is no defined protocol regarding the addition of medical therapy after a conservative surgical procedure. Some authors reported the appearance of signs of a persistent ectopic pregnancy after conservative laparoscopic

tubal surgery and the subsequent resolution of symptoms and normalization of hCG levels after treatment with methotrexate⁽¹⁸⁾. Accordingly, we decided to administer a single-dose MTX injection to reduce the risk of post-salpingostomy ectopic pregnancy persistence.

CONCLUSIONS

BTP represents an intriguing condition that has been seldom reported in medical literature. Surgeons should always keep in mind the possibility of BTP, especially when patient has one or more risk factors in her history. Failure in the pre-operative evaluation of the adnexa contralateral to the identified tubal pregnancy, may give rise to a suspicion. Counseling is very important in these cases and, consequently, the therapeutic approach must be tailored for every patient.

REFERENCES

- 1) Al-Quraan GA, Al-Taani MI, Nusair BM, El-Masri A, Arafat MR, Khateeb MM. **Spontaneous ruptured and intact bilateral tubal ectopic pregnancy.** Eastern Mediterranean Health Journal 2007;13:967-72.
- 2) Aanesen A, Flam F. **Bilateral tubal pregnancy following in vitro fertilization and transfer of two embryos.** European Journal of Obstetrics & Gynecology and Reproductive Biology 1996;64:235-6.
- 3) Hoopmann M, Wilhelm L, Possover M, Nawroth F. **Heterotopic triplet pregnancy with bilateral tubal and intrauterine pregnancy after IVF.** Reprod Biomed Online. 2003;6:345-8.
- 4) Li W, Wang G, Lin T, Sun W. **Misdiagnosis of bilateral tubal pregnancy: a case report.** Journal of medical case reports 2014;8:342-5.
- 5) Lee JD, Chang SY, Chang MY, Lai YM, Soong YK. **Simultaneous bilateral tubal pregnancies after in vitro fertilization and embryo transfer: report of a case.** J Formos Med Assoc. 1992;91:99-101.
- 6) Altinkaya SO, Ozat M, Pektas MK, Gungor T, Mollamahmutoglu L. **Simultaneous bilateral tubal pregnancy after in vitro fertilization and embryo transfer.** Taiwan J Obstet Gynecol. 2008;47:338-40.
- 7) Ryan MT, Saldana B. **Bilateral Tubal Ectopic Pregnancy: A Tale of Caution.** Academic Emergency Medicine 2000;7:1160-3.
- 8) Marasinghe JP, Condous G, Amarasinghe WI. **Spontaneous bilateral tubal ectopic pregnancy.** Ceylon Med J. 2009;54:21-2.
- 9) Wali AS, Khan RS. **Spontaneous bilateral tubal pregnancy.** J Coll Physicians Surg Pak. 2012;22:118-9.
- 10) Tabachnikoff RM, Dada MO, Woods RJ, et al. **Bilateral tubal pregnancy: a report of an unusual case.** J Reprod Med. 1998;43:707-9.
- 11) Amine BH, Haythem S. **Extra-uterine twin pregnancy: case report of spontaneous bilateral tubal**

- ectopic pregnancy.** Pan Afr Med J. 2015;20:435.
- 12) Fishback HR. **Bilateral simultaneous tubal pregnancy.** Am J Obstet Gynecol. 1939; 37:1035-7.
- 13) Norris S. **Bilateral simultaneous tubal pregnancy.** Can Med Assoc J. 1953;68:379-81.
- 14) Xiromeritis P, Margioulas-Siarkou C, Miliaras D, Kalogiannidis I. **Laparoscopic excision of coexisting left tubal and right pseudotubal pregnancy after conservative management of previous ectopic pregnancy with methotrexate: an unusual clinical entity.** Case Rep Surg. 2015;2015:645826.
- 15) Cucinella G, Rotolo S, Calagna G, Granese R, Agrusa A, Perino A. **Laparoscopic management of interstitial pregnancy: the "purse-string" technique.** Acta Obstet Gynecol Scand. 2012;91:996-9.
- 16) Idotta R, Tripodi A, Scopelliti P. **Bilateral, tubal pregnancy treated with conservative endoscopic surgery.** Clin Exp Obstet Gynecol. 2001;28:107-8.
- 17) Sommer EM, Reisenberger K, Bogner G, Nagele F. **Laparoscopic management of an unrecognized spontaneous bilateral tubal pregnancy.** Acta Obstet Gynecol Scand. 2002;81:366-8.
- 18) Andrews J, Farrell S. **Spontaneous bilateral tubal pregnancies: a case report.** J Obstet Gynaecol Can. 2008;30:51-4.
- 19) Liao CY, Ding DC. **Laparoscopic management of spontaneous bilateral tubal pregnancies.** J Minim Invasive Gynecol. 2009;16:247.
- 20) Mathlouthi N, Jellouli MA, Temime RB, Makhlof T, Attia L, Abdellatif C. **Spontaneous and simultaneous bilateral tubal pregnancy.** La Tunisie Medicale. 2012;90:582
- 21) Rani VRS, Puliyath G. **Viable intrauterine pregnancy after spontaneous bilateral tubal ectopics in a multiparous woman: a case report.** J Med Case Rep. 2013;7:159.