Spontaneous Bilateral Tubal Pregnancies: A Case Report

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Abstract

Background: Bilateral tubal pregnancies in the absence of preceding induction of ovulation are a rare occurrence, with an estimated incidence of 1 in 725 to 1 in 1580 ectopic pregnancies. They are usually diagnosed at the time of surgery.

Case: We report a case of spontaneous bilateral tubal pregnancies diagnosed intraoperatively. Conservative laparoscopic tubal surgery was used to treat the ectopic pregnancies. The patient returned with signs of a persistent ectopic pregnancy and was subsequently treated with methotrexate with resolution of symptoms and normalization of hCG levels.

Conclusion: The diagnosis of bilateral tubal pregnancy is usually made intraoperatively. This underscores the importance of identifying and closely examining both tubes at the time of surgery, even in the presence of significant adhesive disease.

Background

The reported prevalence of extrauterine pregnancy in the Canadian population is approximately 13.8 per 1000 reported pregnancies. It remains the most common life-threatening emergency in early pregnancy, accounting for approximately 10% of all maternal deaths in industrialized nations. The incidence of ectopic pregnancy has been reported to be increasing in many countries in recent years as a result of a number of factors: increases in rates of sexually transmitted infections that damage the fallopian tubes, the use of antibiotic treatments for pelvic inflammatory disease rather than surgical removal of the tubes, more accurate methods for early detection of ectopic pregnancy, increased use of assisted reproductive technologies, and increased rates of tubal sterilization. The incidence of ectopic pregnancy in the United States has increased from 4.5 per 1000 pregnancies in 1970 to 19.7 per 1000 pregnancies in 1992. Conversely, the rate in Canada has decreased in the past decade from 16.4 per 1000 pregnancies in 1991–1992 to 13.8 per 1000 pregnancies in 1999–2000. Ectopic pregnancy still accounted for approximately 8% of direct maternal deaths between 1997 and 1999.

Bilateral tubal pregnancies in the absence of preceding induction of ovulation are an extremely unusual occurrence and are thought to represent the rarest form of extrauterine pregnancy. More common are twin pregnancies in the same tube and heterotopic pregnancies. Fishback was the first to report the criteria for diagnosing an authentic case of bilateral simultaneous extrauterine pregnancy. He declared that there should be a description of the fetuses or fetal parts as well as of placental material. Norris claimed that microscopic identification of chorionic villi in each tube should suffice.

The incidence of simultaneous bilateral tubal pregnancies has been reported to range from 1 per 725 to 1 per 1580 ectopic pregnancies. This is thought to correspond to an occurrence of one per every 200 000 live births.

Key Words: Pregnancy, ectopic, tubal, bilateral, pregnancy complications, salpingostomy

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There has been an increase in the number of published case reports of bilateral tubal pregnancies associated with the increasing use of assisted reproductive technologies.\(^9,10\) However, without iatrogenic ovulation induction, simultaneous bilateral tubal pregnancy remains an atypical event.

**THE CASE**

A 25-year-old gravida 3, para 0 woman was referred to the gynaecology service with an approximate gestational age of 9 weeks and 2 days. Presenting complaints included vaginal bleeding and intermittent lower abdominal cramping; the patient was hemodynamically stable. The initial level of serum $\beta$-hCG was 24,242 IU/L. A transvaginal pelvic ultrasound examination revealed an empty uterus with a right adnexal mass measuring 4.3 x 2.3 cm. The patient’s past gynaecologic history included two previous therapeutic abortions. She had no history of sexually transmitted infections or previous abdomino-pelvic surgery. She was otherwise healthy.

The patient was counselled concerning the possibility of an ectopic pregnancy, and informed consent for laparoscopic exploration with the possible need for salpingostomy or salpingectomy was obtained.

At laparoscopy, a small amount of bleeding was noted in both the anterior and posterior cul-de-sacs. The right tube contained a mass measuring approximately 2 x 3 cm; its appearance was consistent with an ectopic pregnancy. The left tube was then found to contain a mass consistent with an ectopic pregnancy (Figure 1).

We considered it possible that the patient had bilateral spontaneous ectopic pregnancies and decided to perform bilateral salpingostomies. After linear salpingostomy a small conceptus with visually identifiable fetal parts was retrieved from the left fallopian tube (Figure 2). This appeared to be a fairly advanced gestation for an ectopic pregnancy. Linear salpingostomy of the right tube produced products resembling hemorrhagic tissue typically found with ectopic pregnancies. There were no complications from the surgery, and the patient recovered well. She was counselled that she was at high risk of subsequent ectopic pregnancy and advised to seek medical attention early in future pregnancies. She was also instructed to have serum $\beta$-hCG levels monitored weekly until they returned to normal.

The pathology report confirmed the diagnosis of spontaneous bilateral tubal pregnancies, showing blood clot admixed with chorionic villi in the tissue removed from the right tube. The tissue obtained from the left tube showed multiple fragments of fetal tissue, including the vertebral column, neurological structures, liver, intestine, umbilical cord, and chorionic villi.

The patient was readmitted approximately four weeks later with recurrent lower abdominal pain. Her serum $\beta$-hCG level was 556 IU/L. Since her serum $\beta$-hCG values were not declining rapidly enough to suggest spontaneous resolution of the ectopic pregnancy, she was given an intramuscular injection of methotrexate 50 mg/m\(^2\). Her pain resolved, and her serum $\beta$-hCG levels declined to an undetectable level.

**DISCUSSION**

Simultaneous bilateral ectopic pregnancy is the rarest form of extraterine gestation. We report a case that meets the criteria outlined by Fishback\(^7\) and Norris\(^5\): fetal tissue and chorionic villi were demonstrated on histopathological examination of the tissue obtained from each tube.

Fishback was the first to conduct a comprehensive review of the medical literature, and he established criteria to validate a diagnosis of simultaneous bilateral tubal pregnancy. He reported a series of 76 patients fulfilling the requisite criteria.\(^7\) Edelstein reviewed the English-language literature in 1989 and found a further 22 cases.\(^8\) A literature search of
English-language journals on Medline reveals 45 further case reports since Edelstein’s review in 1989. Of these, 17 are associated with various treatments for infertility and assisted reproductive techniques, and 28 cases are listed as spontaneous.

Several theories have been postulated in an attempt to explain the occurrence of bilateral tubal pregnancies. Foster stated that bilateral tubal gestation requires multiple ovulations to occur, the oocytes to be fertilized, and the oocytes to implant at sites of tubal damage.11 Another possible etiology is transperitoneal migration of trophoblastic cells from one tube to another, which is a plausible explanation for the finding of fetal tissue in one tube and only villi in the other tube reported by Tabachnikoff et al.12 However, this discrepancy may be the result of unequal growth of the two gestations, as has been reported in several previous cases.6,8,13,14 Tubal pregnancies are known to develop and grow abnormally; furthermore, arrests of development have been characterized in ectopic pregnancies.10,12

Superfetation, another possible etiology, implies fertilization and development of a second oocyte when a woman is already pregnant.12 This is considered to be an extremely rare event in humans and is inherently difficult to prove.14 The diagnosis is usually suspected when severe growth discrepancy is apparent in a multiple pregnancy.15 Several case reports have been published suggesting superfetation as an explanation for consistent growth discordance in twin gestations diagnosed on early ultrasound.15–17

Previous studies have demonstrated that serum estradiol, progesterone, and serum β-hCG levels are all lower in ectopic gestations than in normal intrauterine pregnancies.18,19 Whether or not the lower hormone levels observed in ectopic gestations are insufficient to inhibit a subsequent ovulation is not known. Urman and McComb20 reported evidence in support of superfetation in a patient who had an ongoing ectopic pregnancy. The patient had a documented rise in mid-cycle basal body temperature, suggesting ovulation occurred while she carried a tubal pregnancy.20 Whether or not this occurred in our patient is unknown. However, this may explain the growth discrepancy observed between the two gestations.

A further explanation may be that the second tubal pregnancy occurred after the demise of the first. If the first tubal gestation spontaneously aborted or was in the process of aborting, a second ovulation may have occurred during this time period, resulting in the finding of bilateral tubal pregnancies. Kobayishi et al. reported a similar case, in which an intrauterine pregnancy occurred during an incomplete abortion of an extrauterine gestation.21

Most patients with bilateral tubal pregnancies present similarly to those with a unilateral ectopic pregnancy and have similar risk factors.22,23 The most frequent findings are the triad of amenorrhea, vaginal bleeding, and abdominal pain.4,8,22 Levels of serum β-hCG and the discriminatory zone are not reliable for patients with bilateral disease.24 Ultrasound also has not been useful in the identification of...
bilateral tubal gestations. Consequently, the diagnosis of bilateral tubal pregnancy is usually made intraoperatively. This underscores the importance of identifying and closely examining both tubes at the time of surgery, even in the presence of significant adhesive disease.

Various treatments have been employed with bilateral tubal pregnancies. These range from the extreme of a total abdominal hysterectomy with bilateral salpingo-oophorectomy to the conservative approach, with laparoscopic techniques involving salpingectomy or salpingostomy. The first case report of laparoscopic treatment was in 1995. Since then, there have been several reports of treatment with both laparoscopic salpingectomy and salpingostomy. There are also several case reports of failure to diagnose bilateral tubal pregnancy at the time of initial surgery. In these instances, the patients returned with increased symptoms or rupture of the contralateral tube. The diagnosis of bilateral pregnancy was hence made at the time of subsequent surgery.

There are no case reports of primary medical treatment with methotrexate for this complication. However, since the diagnosis of bilateral tubal pregnancy is usually made at the time of surgery, it is plausible that there may be cases in which clinicians are unaware that medical treatment of patients has resulted in successful management of bilateral tubal pregnancy. Walter reported a case of bilateral chronic and acute tubal pregnancies following failed treatment with methotrexate for a previous ectopic pregnancy.

In our case, the patient presented with symptoms and clinical findings consistent with a presumed diagnosis of unilateral ectopic pregnancy. Fortunately, the pregnancies were unruptured and the patient was hemodynamically stable. We decided to proceed with conservative tubal surgery. Treatment with conservative surgery has been effective in similar situations, and there have been at least two instances of successful intrauterine pregnancy following conservative treatment. Nonetheless, the fact remains that these patients are at high risk for recurrent ectopic pregnancy.

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The woman whose story is told in this case report has provided signed permission for its publication.

REFERENCES