Spontaneous Triplet, Tubal Ectopic Gestation

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Only six cases of spontaneous, unilateral, triplet ectopic gestations have previously been reported. We now present a seventh case. The patient's prior obstetrical history was significant for a term stillbirth and a term cesarean section for breech. Quantitative ßhCG was normal for gestational age; however, the increased trophoblastic mass of an inappropriately implanted multiple gestation may produce sufficient ßhCG to mimic an intrauterine singleton gestation.

Resolution was achieved via salpingostomy. This case is significant for being spontaneously conceived and not the result of assisted reproductive technologies. Furthermore, this case supports an association between prior cesarean section and ectopic gestation.

Key words: triplet ■ ectopic gestation

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In 1992, the incidence of ectopic gestations was 19.7 per 1,000 pregnancies in the United States.[1] The prevalence of all risk factors for ectopic gestations such as tubal pathology, sterilization, prior tubal ectopic gestation, infertility and previous genital tract infections are increasing.[1,2] Ectopic gestations are now the leading cause of first-trimester maternal deaths, and 90% of these are due to hemorrhage.

Ninety-eight percent of ectopic gestations are located in the fallopian tube, with the most common site being the ampullary portion. Ectopic gestations may be simultaneous with intrauterine gestations, but it is estimated that heterotopic gestations occur in only 1:3,889 gestations. Multiple ectopic gestations may be due to simultaneous bilateral ovulation, or superfetation with or without trans peritoneal migration.

Almost 100 cases of unilateral ectopic twin gestations and many bilateral twin ectopic gestations (an incidence of up to 1:750) have been reported, but only six cases of spontaneous, unilateral, triplet ectopic gestations have been documented.[3,4] Here, we report the seventh case of a spontaneous, unilateral, triplet, tubal ectopic gestation without any contributory gynecologic factors.

CASE REPORT

B.D., an obese 34-year-old woman, gravida 4 para 3002, presented at eight weeks and six days' estimated gestational age by last normal menstrual period to an ambulatory gynecology clinic. She complained of a four-day history of disabling, severe, intermittent, sharp, crampy, nonradiating "gas pains" bilateral in the lower quadrants and without temporal or aggravating factors. She noted an unusual menstrual period a month prior (4 + 6/7 weeks' estimated gestational age), lasting three days followed by a positive urine pregnancy test and several days of vaginal spotting. She had a noncontributory gynecologic history. She had twice delivered vaginally at term, including one stillborn followed by a primary cesarean section at term for breech.

On physical exam, B.D. was not in acute distress. The abdomen was soft and nondistended with greater right than left lower quadrant tenderness to palpation, lacking rebound or guarding. Pelvic examination lim-

ited by a BMI of 36 showed minimal blood at the cervical os, and a mid-position 6–8-week size uterus.

A complete blood count and quantitative b-human chorionic gonadotrophin (ßhCG) were drawn. The initial plan was to perform a pelvic ultrasound if the quantitative ßhCG was sufficient for visualization of a gestation. When she presented early the following morning with increasingly severe right abdominal pain and adnexal tenderness to palpation on examination, her initial labs had resulted: ßhCG of 73,954 mIU/mL and a hematocrit of 33.7%. Abdominal ultrasound showed a large right proximal tubal ectopic gestation with a crown rump length and sac size consistent with nine weeks' estimated gestational age. A significant amount of free fluid was present in the cul de sac. A pseudosac was seen in the uterine cavity.

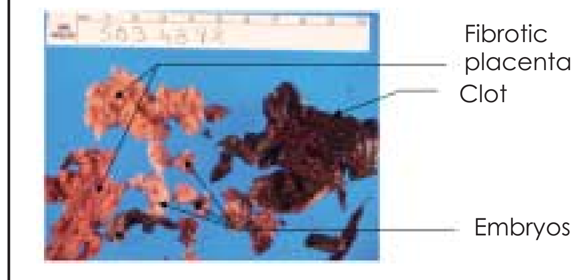
At surgery, hemoperitoneum of approximately 350 cc and a grossly dilated, unruptured, right fallopian tube approximately 4 cm in diameter were seen. A right linear salpingostomy was performed over the antimesenteric portion of the tube via exploratory laparotomy. Three fetuses (Figure 1) in two gestational sacs were extracted from the fallopian tube, which was adherent to the right ovary. The incision was left open and the edge oversewn with a simple running stitch of 4-0 braided, coated polyglactin suture. B.D. was discharged home on postoperative day 1 (POD 1) with a ßhCG of 16,942 mIU/mL and a hematocrit of 27.2%. On POD 5 follow-up, the ßhCG was 2,338 mIU/mL.

DISCUSSION

Cesarean section in a multipara may be a significant risk factor for subsequent ectopic gestation, with a relative risk of 1.28 in Hemminki and Merilainen's retrospective cohort[5] and so should be considered along with the aforementioned historical risk factors when evaluating women whose presenting signs and symptoms are suggestive of ectopic gestation. However, the literature survey is equivocal; formal meta-analysis may clarify the association, if any, of prior cesarean section and ectopic gestation. Bastianelli et al. showed a doubling of risk of ectopic gestation with prior cesarean section based on 213 cases and 213 controls, whereas Butts et al. (with 203 cases and 61 controls) and Kendrick et al. (using 138 cases and 842 controls) show no association between ectopic gestation and prior cesarean section.[6-8] Combined these four studies give an approximately 1.3-fold increased risk of future ectopic gestation following cesarean section. This is significant for the case presented, as most multiple ectopic gestations are associated with usage of assisted reproductive technologies, intrauterine devices or prior tubal surgery. Thus, prior cesarean section would be B.D.'s only potential risk factor for an ectopic gestation.

Early ultrasonography may eliminate other differential diagnoses: threatened, spontaneous and incomplete abortions. Ultrasonography has replaced culdocentesis to confirm the presence of free fluid in the cul de sac and abdominal cavity found in up to 96.2% of ectopic gestations. Ultrasonography permits visualization of a gestational sac identified by a round hypoechogenic fluid collection surrounded by a hyperechogenic rim of the trophoblast. Pseudogestational sacs are nonspecific collections of fluid due to decidualization of the endometrium, found in up to 9.4% of ectopic gestations.

Figure 1. Products of conception



Trophoblast-produced human chorionic gonadotrophin (hCG) reaches detectable serum levels within 10 days of fertilization. The quantitative ßhCG doubles every 48–72 hours. Normally increasing ßhCG may occur in up to 7% of ectopic gestations, with 49% of ectopic gestations having a declining ßhCG and 44% having smaller increases in quantitative ßhCG. Given the potentially greater trophoblastic mass of higher-order multiple ectopic gestations, such gestations could exhibit the normal doubling of ßhCG every 48–72 hours that occurs in 7% of ectopic gestations.

Given increasing numbers of multiple gestations via assisted reproductive technologies, it may easily be forgotten that higher-order multiple gestations can, and do, also occur spontaneously. This case is further evidence that higher-order ectopic gestations are naturally occurring and may present atypically in comparison to singleton ectopic gestations.

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