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Case Report

Spontaneous Heterotopic Pregnancy after Four Previous Caesarean Sections: Successful Salpingectomy and Intra-Uterine Term Pregnancy

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Heterotopic pregnancy is rare under natural circumstance (conception). We report the first case of heterotopic pregnancy in spontaneous conception with four previous caesarean sections and ruptured tubal ectopic with resultant successful normal intra-uterine pregnancy delivered at term after salpingectomy. A 32-year-old gravida 5 para 4+0 with 3 living children, with 4 previous caesarean sections, presented to the Accident and Emergency unit of Nnamdi Azikiwe University Teaching Hospital, Nnewi at a gestational age of 9 weeks + 2 days, with recurrent vaginal spotting of 2 weeks duration, lower abdominal pain as well as nausea and vomiting of 3 days duration. Initial transvaginal ultrasound scan confirmed intrauterine gestation at 9 weeks of gestational age, with right adnexal mass which was misdiagnosed as corpus luteum. Later, a diagnosis of heterotopic pregnancy was made and she had laparotomy with right salpingectomy. The intrauterine pregnancy was continued till term. She subsequently had elective repeat caesarean section with the outcome of a live male baby of Apgar scores of 8 and 10 in the first and the fifth minutes respectively, and birth weight of 4.3 kg. This demonstrates that spontaneous heterotopic pregnancy can occur after four previous caesarean sections, and that the presence of corpus luteum in early ultrasound scan should raise the suspicion of possible heterotopic pregnancy. Early detection and prompt intervention is necessary for the safety of both the mother and the normal intrauterine gestation.

Keywords: Heterotopic pregnancy, spontaneous conception, caesarean section, hemoperitoneum, ultrasound

Heterotopic pregnancy refers to the presence of simultaneous pregnancies at two different implantation sites. Most often a combination of intrauterine and ectopic pregnancies are encountered, rather than two ectopic ones (1, 2).

Estimated world-wide incidence of heterotopic pregnancy in a spontaneous conception is 1 in 30,000 (2). In contrast, ectopic pregnancy occurs in about 2% of all pregnancies. However, with the advent of widespread use of ovulation induction drugs such as clomiphene and other assisted reproductive techniques, the incidence has now been thought to be as high as 1 in 3900 pregnancies (3-5), and even higher if there has been pre-existing tubal damage. In the USA, analysis of assisted reproductive techniques pregnancies from 1999-

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2002 showed an incidence of heterotropic pregnancy at 1.5 per 1000 (6).

The risk of ectopic pregnancy increases significantly after pelvic inflammatory disease; mainly chlamydial infections, widespread use of intrauterine devices, microsurgical repair of tubal damage caused by pelvic inflammatory disease (7). Incidentally, the woman has none of all these apart from the surgeries.

Delay in diagnosing the condition and failure to proceed quickly with the requisite anesthesia and surgery can compromise both maternal health and survival of the intra-uterine fetus.

Here we present a case of heterotopic pregnancy in spontaneous conception with four previous caesarean section with ruptured tubal ectopic and hemoperitoneum at Nnamdi Azikiwe University Teaching Hospital Nnewi, Anambra State Nigeria.

Case report

Mrs. BI, was a 32-year-old gravida 5 para 4+0 with 3 living children. She presented to our Accident and Emergency unit at Nnamdi Azikiwe University Teaching Hospital, Nnewi, Nigeria on 26 May 2015 with two weeks history of recurrent vaginal spotting, neither passage of vesicles nor fainting spells or dizziness. She also complained of lower abdominal pain of one week duration which was more at the right iliac fossa and suprapubic region. She described the pain as sporadic, which was exacerbated by movement and resolving with rest. There was associated nausea and vomiting. It was a spontaneous conception and there was no past history of abortion, infertility, pelvic infections, no previous history of ectopic pregnancy, reconstructive surgery, or hormonal contraceptives use. She knew she was in a gravid state. On examination, her general condition was stable, not pale, afebrile, anicteric, not dehydrated, blood pressure- 120/80 mmHg, pulse rate- 70 beats per minute, respiratory rate-18 cycles per minute. There was tenderness over the hypogastrium and right iliac fossa.

Vaginal speculum examination revealed scanty blood in the vaginal vault and closed cervical os. The uterine size could not be assessed, due to generalized pelvic tenderness. Hemoglobin concentration was 10 g/dl. Trans-vaginal ultrasound performed the same day revealed a viable intra-uterine fetus at 9 weeks + 2 days gestational age, with uterine fibroid longest measuring 32 mm x 29 mm, concurrently the right adnexum had a mass measuring about 60 mm x 40 mm (which was mistaken to be corpus luteum cyst). A diagnosis of corpus luteum cyst was made. She was counseled, reassured and placed on bed rest.

However, symptoms persisted which necessitated her re-evaluation 2 days after her initial presentation. The blood pressure was 110/70 mmHg, heart rate was 80 beats per minute and respiratory rate was 28 cycles per minute. Abdominal examination elicited right lower quadrant tenderness, with rebound and guarding. A repeat trans-vaginal ultrasound further revealed a viable intra-uterine gestation with right adnexal mass and hemoperitoneum. At this point, a diagnosis of heterotopic pregnancy with ruptured right tubal ectopic pregnancy was entertained. She was counselled for an emergency laparotomy and the couple gave consent.

During the operation, hemoperitoneum of about 800 ml was found together with a ruptured right tubal (ampullary) ectopic pregnancy. The uterus was carefully handled, left tube and both ovaries were normal. Suction of hemoperitoneum and right sided salpingectomy was carried out. The patient recovered well and was discharged home on 8th postoperative day. A repeat ultrasound showed that the intra-uterine gestation was still viable. Subsequently, she was placed on progesterone for pregnancy support. Histologic examination of specimen sample confirmed right ectopic gestation.

Further abdominal ultrasound (28 July 2015) 8 weeks after the laparotomy revealed a viable intra-uterine singleton fetus with crown rump length of 92 mm at a gestational age of 15 weeks + 3 days. She

received antenatal care and the pregnancy continued to term. On 15 December 2015, she had an elective lower segment caesarean section with an outcome of a live male baby that weighed 4.3 kg and Apgar's scores of 8 and 10 in the first and fifth minutes, respectively. She had left tubal ligation intraoperatively. The patient recovered well and was discharged home on postoperative day 8. She signed permission of this publication.

Discussion

Although, cases of spontaneous heterotopic pregnancies have been reported recently in Nigeria (8, 9), our case is unique, because, to the best of our knowledge this is the first report of spontaneous heterotopic pregnancy occurring in a women after four caesarean sections.

The clinical presentation of heterotopic pregnancy mimics that of ectopic pregnancy in other site, and symptoms of threatened miscarriage. Mostly, heterotopic pregnancies present with abdominal pain, vaginal bleeding, adnexal mass, peritoneal irritation, etc. (2). This patient had all these symptoms.

Patients are often diagnosed at a more advanced gestational age in comparison with isolated tubal ectopic pregnancy because when an intrauterine pregnancy is observed at ultrasound, the possibility of an additional ectopic one is not generally considered. As a result of misdiagnosis, there is high risk of rupture at presentation (10). On average, the gestational age at diagnosis is 8 weeks ± 3 weeks (10). The woman under review was diagnosed at 9 weeks of gestation.

All women of reproductive age with abdominal pain, vaginal bleeding should undergo pregnancy test. If positive, localization of the pregnancy is made with ultrasound. Signs suggestive of heterotopic pregnancy are complex adnexal mass or fluid in the pelvis, in the presence of an intrauterine pregnancy. If clinicians have high index of suspicion for heterotopic pregnancy after visualizing an intrauterine pregnancy, the ectopic pregnancy may be labeled as corpus luteum cyst. Advanced ectopic pregnancy containing a yolk sac or fetal poles with cardiac activities makes this diagnosis easier to make (4). Unfortunately, the diagnosis was missed initially until the patient had tubal rupture. Beta human chorionic gonadotropin assay is not useful as it primarily reflects the intrauterine pregnancy. The differential diagnoses include threatened miscarriage, ruptured corpus luteum cyst, appendicitis, nephrolithiasis, etc.

The treatment of heterotopic pregnancy should utilize the less invasive therapy in order to preserve the concomitant intrauterine pregnancy. Laparoscopy is preferred over laparotomy for the treatment of heterotopic pregnancy because of its favorable profile for less bleeding, pain, hospitalization, recovery time and cost (10, 11). For an extra-uterine gestation in the tubal location, salpingectomy is indicated over salpingostomy (10). The latter may result in incomplete removal and persistent ectopic pregnancy, which would be impossible to identify with serial beta hCG level given the existing intrauterine pregnancy. For hemodynamically unstable patients, laparotomy is referred (11, 12). Our patient had laparotomy with right total salpingectomy. There is no place for systemic chemotherapy if the intrauterine pregnancy is viable. (11). Also, laparoscopic surgery has little or no role as the woman had history of four previous caesarean sections.

The present case demonstrates that spontaneous heterotopic pregnancy can occur after four previous caesarean sections, and that the presence of corpus luteum in early ultrasound scan should raise the suspicion of its possibility. Early detection and prompt intervention is necessary for the safety of both the mother and the normal intrauterine gestation.

Conflict of interest

The authors declare that they have no competing interest.

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