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CASE REPORT

Heterotopic pregnancy with natural conception; a rare event that is still being misdiagnosed: a case report

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Key Clinical Message

Simultaneous presence of intra- and extra-uterine pregnancies have been known as heterotopic pregnancy (HP); the condition which is extremely rare with natural conception. Our aim is to increase the obstetrician's awareness to increase the level of positive outcome in such rare events.

Keywords

Acute abdomen in pregnancy, extrauterine pregnancy, heterotopic pregnancy.

Introduction

Heterotopic pregnancy is a rare condition that is characterized by presence of simultaneous intra- and extra-uterine pregnancy [1]. It mostly happens with known risk factors notably assisted reproductive techniques (ART), infertility treatment, and previous history of pelvic inflammatory diseases (PID) [2].

Patients with known risk factors are usually kept under strict follow-up schedules that include booking an antenatal scan with subsequent early detection of any possible HP.

The diagnostic challenge still remains in those who present with HP without identifiable risk factors and with spontaneous pregnancy, and those who usually attend first with complicated HP, are usually overlooked.

We want to report our experience of HP after spontaneous pregnancy, and unluckily misdiagnosed with ultimately, loss of the intrauterine pregnancy. Our hope is to raise awareness for such a rare event to ensure better patient outcomes.

Case Presentation

A 38-year-old pregnant woman G7 P3 (Ab3) came to our antenatal emergency unit with abdominal pain that was in the right lower quadrant, aching, continuous in nature, and associated with two vomiting episodes of clear fluid. No dysuria, vaginal bleeding, discharge or fever. Her last menstrual period (LMP) was 9 weeks previously, and she had no prior history of ectopic pregnancy, artificial reproductive techniques (ARTs), pelvic inflammatory disease (PID), or infertility treatment.

Upon examination, the patient appeared stressed from pain, vital signs were within normal limit, and an abdominal examination revealed tenderness at right iliac fossa and positive rebound tenderness. Nothing abnormal was noted by vaginal examination.

Routine work up was initially done that revealed, hemoglobin level: 11.8/dL, white cell count: 13.19/mm^3, and *B*-HCG: 45,200 mIU/mL. Transabdominal ultrasound showed gravid uterus with living fetus corresponding 6 + 5 weeks (Fig. 1) and right adnexal mass (10 × 8 × 7 cm) with mild to moderate free fluid (Fig. 2). Our radiologist and obstetrician got a false assurance as they saw a viable intrauterine fetus by ultrasound and kept the possibility of the HP in the bottom of the differential diagnosis list.

Signs of acute abdomen and clinical picture of suggestive complicated appendicitis warranted urgent intervention. The patient and her husband were informed of all

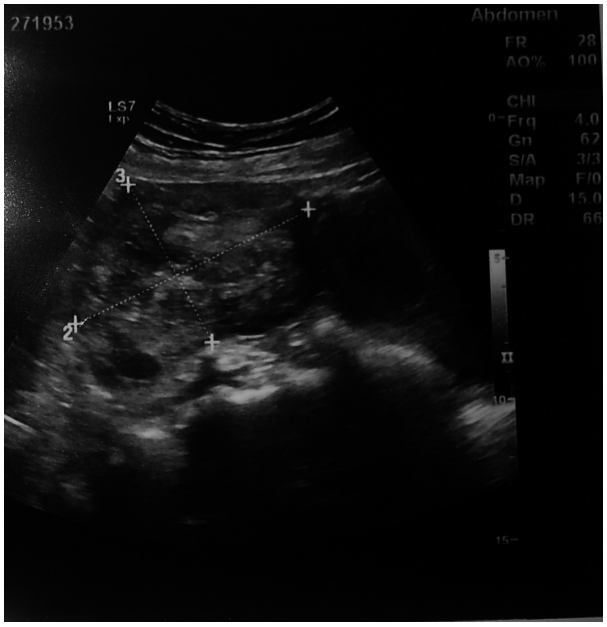

Figure 1. Transabdominal ultrasound showing right adnexal mass.

the benefits and risks of such a procedure that include the possibility of miscarriage from general anesthesia and surgery.

After an initial grid iron (McBurney's) incision and as the peritoneum was opened, there was hemoperitoneum, and the right fallopian tube was found to be ruptured (Fig. 3). The incision was extended by OBE/GYNAE team and a total right salpingectomy was performed. Also, appendectomy was done that was grossly normal (Fig. 4).

Histopathology confirmed presence of chorionic villi suggestive of HP and normal appendix.

The patient was discharged with uneventful postoperative period but unfortunately she return after 12 days suffering incomplete miscarriage that was managed by medical termination.

Comment

Our reported case is a ruptured right tubal HP after spontaneous pregnancy. The case was initially misdiagnosed as complicated appendicitis and ended with miscarriage of the intrauterine one.

Heterotopic pregnancy is a rare event with a reported incidence in the general population of 1 in every 7000 pregnancies. The incidence has increased due to widespread use of ART and history of PID to approximately 1 in 100 pregnancies. It's extremely rare among patients who conceive naturally, <1 in 30,000 [3, 4]. In a series of 13 cases of HP, one case only was after spontaneous pregnancy, six after ART and six after infertility treatment [5].

The fallopian tube is the most frequent reported site (72%) for extrauterine pregnancy, as was our case. Also,

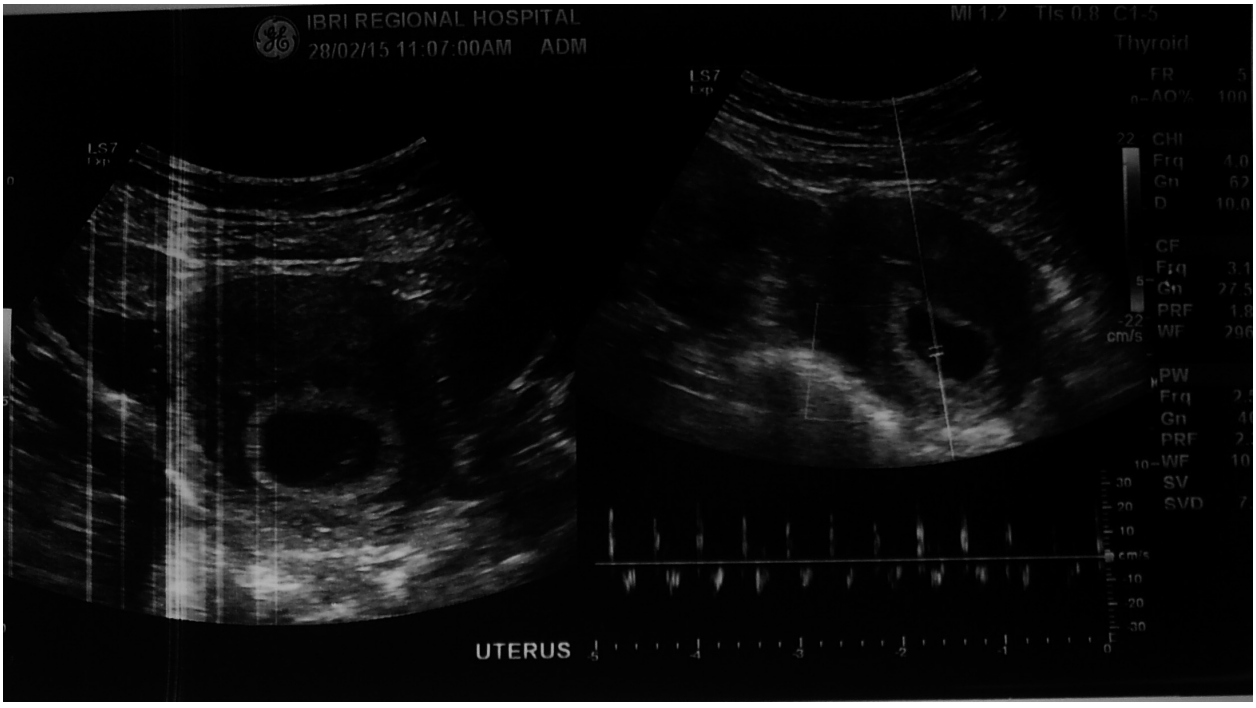

Figure 2. Transabdominal ultrasound showing intrauterine gestational sac with a live fetus.

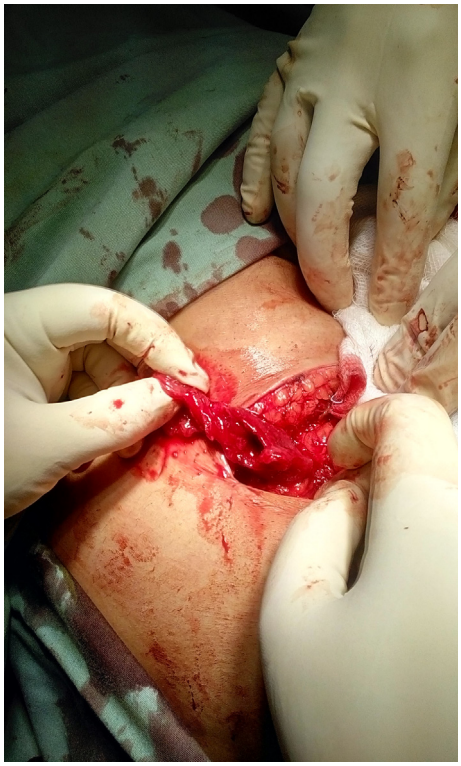

Figure 3. Intraoperative finding; a ruptured right fallopian tube.

ovarian and cervical sites [6, 7], and spontaneous triple HP with two yolk sacs in one tube were reported [8].

Accurate and early diagnosis of HP is challenging and still remains a diagnostic and therapeutic challenge to practitioners despite the increased medical knowledge and use of improved reproductive technologies [2, 3].

Few modalities may help in the HP diagnosis of HP, notably *B*-HCG and transvaginal ultrasound. Serial samples of *B*-HCG measurements are often difficult to interpret because of intrauterine pregnancy, and cause the *B*-HCG concentration to increase appropriately. The serum level of *B*-hCG in our case was 45.200 mIU/mL, which is around the normal level for intrauterine pregnancy at this period of gestation.

Routine transvaginal ultrasound detected only 56% of HP cases who underwent ART [3]. Our case was really a challenging one as she did not have any of the known risk factors of HP, was conceived naturally and had no antenatal visit prior to her emergency presentation. Moreover, serum *B*-HCG was around the normal range and transvaginal ultrasound could not help for diagnosis. Missed HP also has reported by Kratschla-Apochal et al. and Gibson Kyle et al. [9, 10]. Nevertheless, it was earlier detected in other series [11, 12].

Medical management can be offered for elective and nonemergent heterotopic pregnancy under some

Figure 4. Gross picture of the surgically removed appendix.

circumstances after accurate diagnosis of the extrauterine gestational sac. Ultrasound-guided vaginal aspiration and *in situ* injection of methotrexate, potassium chloride, or hyperosmolar glucose have been reported [13]. Still, surgical treatment is a good choice for effective resolution of HP. Medical treatment is suitable for selected patients [3].

Immediate intervention remains the only viable option for acute presentation of HP; laparoscopy offers both diagnosis and management and allows for minimal handling of the uterus. Laparoscopy can be considered as a safe option among most of the gynecologists, the outcome of the intrauterine pregnancy is comparable to that obtained with laparotomy [5]. Laparoscopic facility was not available at the time of our case exploration.

Intrauterine fetal outcome after surgical management of HP depends on many factors. Preoperatively early detection of extrauterine pregnancy occurs especially during dating scan and postoperative factors may include short operative time, quality of anesthesia, and minimal uterine manipulation.

Our case ended by loss of the patient's intrauterine pregnancy. However, it was suggested that in those undergoing laparotomy, the loss rate is around 40% of fetuses [14].

Although a rare event, presence of intrauterine pregnancy doesn't eliminate the possibility of HP in pregnant women who present with adnexal mass, even after natural conception. In suspicious cases and with unavailability of laparoscopy, abdominal exploratory incision is recommended. Loss of intrauterine pregnancy may be a deleterious consequence of misdiagnosis.

Clinical key message

Heterotopic pregnancy has mentioned infrequently in the literature, and it seems that physicians have not had full

awareness by such a rare condition. Our case report is a typical example. The patient had heterotopic pregnancy that was misdiagnosed as complicated appendicitis with a delay in diagnosis and intervention that ended by loss of the intrauterine pregnancy days after the exploration. We hope to convey our message to the physicians to alert them to such a rare event that will improve future patient outcomes.

Consent

A written informed consent was taken from the patient for publication of this case. It is available for review from the Journal's Editor-in-Chief.

Acknowledgment

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Conflict of Interest

None declared.

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