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Case Report

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Heterotopic Gestation with a Successful Obstetrical Outcome: A Case Report

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ABSTRACT

Heterotopic pregnancy is a rare condition when at least two pregnancies are present simultaneously at different implantation sites. Such cases may lead to devastating outcomes if nor properly handled.

The patient is a 38-year-old Gravida 6 Para 4 (4014), who presented with severe hypogastric pain, pallor, amenorrhea with positive pregnancy test and a transvaginal ultrasound showing a strong consideration of heterotopic gestation. Emergency exploratory laparotomy for ruptured tubal pregnancy, Left salpingectomy, and evacuation of hemoperitoneum was done, with a bulbously enlarged left fallopian tube with a 2-cm point of rupture seen intraoperatively. The procedure was well tolerated and postoperative course was generally uneventful. Repeat transvaginal ultrasound on hospital day three prior to discharge revealed a live intrauterine gestation at 10 weeks and 1 day.

At 38 weeks and 1 day age of gestation, the patient delivered vaginally to a term, live, baby boy in cephalic presentation, birth weight of 3600 grams, APGAR score of 8, 9 followed by postpartum intrauterine device insertion. The patient was eventually discharged on hospital day 2 without fetomaternal complications.

In conclusion, a successful intrauterine pregnancy with a ruptured heterotopic gestation who underwent surgical intervention though rare is a great possibility. Still, high index of suspicion, complete history and physical examination, sonographic findings in the light of a pregnancy test positive are all imperative to its diagnosis and appropriate management.

Keywords: Heterotopic Pregnancy, Ectopic Pregnancy, Viable Pregnancy

Introduction

Heterotopic gestation is a rare condition, defined as a presence of both intra- and extrauterine pregnancies at the same time [1]. The incidence of a spontaneous heterotopic pregnancy is just 1 in 30,000 spontaneous pregnancies; about 0.08% in all pregnancies. Such coexistent conception with successful live, term intrauterine gestation comes 1 in 60,000 cases or barely 0.0017% [2]. This may be a life-threatening condition if left ignored without proper intervention especially if the extrauterine one ruptures, which necessitates exploratory laparotomy. This procedure may lead to possible morbidity and mortality, thereby decreasing the probability of survival of the coexistent intrauterine gestation.

This study aims to report a heterotopic pregnancy with an intrauterine gestation carried to term and delivered spontaneously after undergoing exploratory laparotomy for tubal pregnancy. Despite almost half of heterotopic gestations leading to unsuccessful outcomes, this case also seeks to document that there are instances of heterotopic pregnancies leading to a successful live birth.

Case Discussion

A 38 year old, Gravida 6 Para 4 (4014), presented to the emergency room of a tertiary government hospital with a sudden onset of non-radiating hypogastric pain of four days duration.

The patient had four previous vaginal deliveries at home. All were carried to term, live, cephalic in presentation, attended by a midwife, without any fetomaternal complications. She had a history of spontaneous miscarriage in 2020, for which, completion curettage was done. Her past medical and family histories were unremarkable. She is a non-smoker, non-alcoholic beverage drinker, and denies illicit drug use.

On physical examination, the patient was awake, alert, not in cardiorespiratory distress, normotensive, tachycardic at 118 beats per minute, afebrile. There were mild signs of pallor and cardiorespiratory distress. Muscle guarding and tenderness were noted on abdominal examination. There was also cervical motion tenderness on pelvic examination. Pertinent findings from the initial work up requested include a positive pregnancy test, a hemoglobin level of 82.0 g/L, and a transvaginal ultrasound (TVS) finding of "early, live intrauterine pregnancy at 9 weeks and 3 days age of gestation, with a complex, thick-walled mass

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in the right adnexa; and a moderate amount of posterior culdesac fluid collection." (Figure 1)

Figure 1: Transvaginal ultrasound pre-operatively. Early, live intrauterine pregnancy at 9 weeks and 3 days age of gestation, with a complex, thick-walled mass in the right adnexa; and a moderate amount of posterior cul-de-sac fluid collection

The patient was admitted as a case of "Gravida 6 Para 4 (4014) Heterotopic pregnancy at 9 weeks and 5 days age of gestation, extrauterine component probably ruptured, advanced maternal age, grandmultigravid;" with the main goal of controlling the internal bleeding form the ruptured extrauterine pregnancy without jeopardizing the intrauterine gestation. An initial of one unit packed Red Blood Cell (pRBC) was transfused and 200 mg progesterone capsule was inserted vaginally to support early pregnancy and prevent miscarriage.

The patient underwent exploratory laparotomy, evacuation of hemoperitoneum, left salpingectomy under spinal anesthesia and was able to tolerate the procedure well. Intraoperatively, the left fallopian tube was noted to be bulbously enlarged, measuring 13 cm x 6 cm x 4 cm with a two-cm point of rupture at the ampullary area. On cut section of the ectopic component, products of conception with blood blots were noted. Uterus was also enlarged to age of gestation with smooth pinkish serosa. The right fallopian tube, and both ovaries were grossly normal (Figure 2). The procedure lasted for 50 minutes with an estimated blood loss of about 1000 mL, 50% of which were evacuated from the hemoperitoneum. There was no immediate post-operative complications. Another one unit of pRBC was transfused to further correct the anemia from acute blood loss. Repeat CBC revealed a hemoglobin count of 107 g/L.

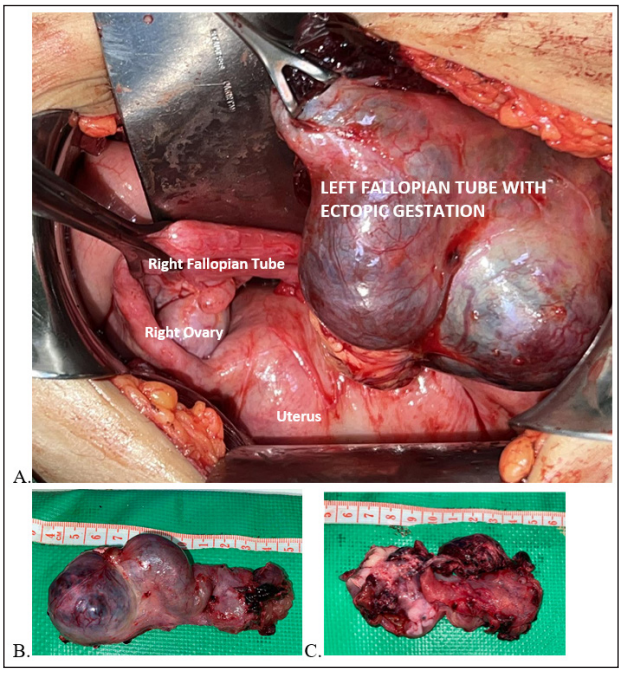


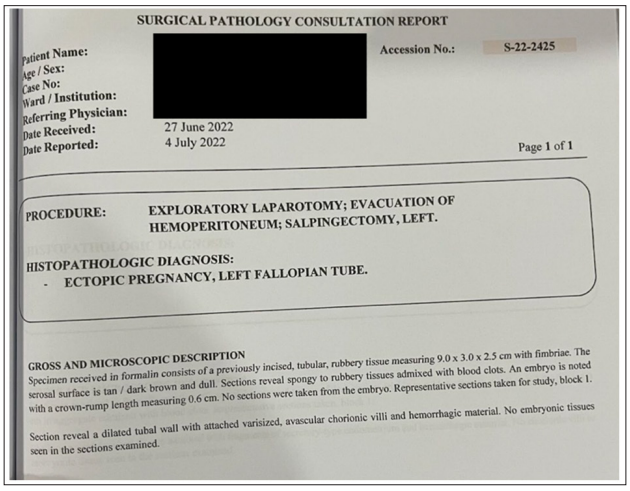
Figure 2: Intraoperative Findings. A. The left fallopian tube was noted to be bulbously enlarged, measuring 13 cm x 6 cm x 4 cm with a 2-cm point of rupture at the ampullary area; B. Ectopic component; C. On cut section, products of conception with blood clots were noted. Uterus was also enlarged to age of gestation with smooth pinkish serosa. The right fallopian tube, and both ovaries were grossly normal.

The patient's immediate post-operative course was unremarkable. recovery at the ward went well. A repeat TVS was done one her third post-operative day with an impression of, "single live intrauterine pregnancy. 10 weeks and 1 day age of gestation by crown to rump length. Good cardiac activity (FHT 174 bpm). No subchorionic hemorrhage. Developing placenta implanted anteriorly. Normal ovaries. Estimated date of delivery on 01/24/2023." She was discharged with the following medications: Folic acid 0.4 mg, and Vitamin B complex capsule once daily as pre-natal vitamins; Cefuroxime 500 mg/cap twice a daily as antimicrobial for 7 days, Paracetamol 500 mg/tablet every 6 hours as needed for pain, and 200 mg micronized progesterone soft gel capsule per vagina and 10 mg/tablet Dydrogesterone three times a day. Biopsy results showed ectopic pregnancy, left fallopian tube.

The patient had regular prenatal check ups at least every four weeks with unremarkable course. A congenital anomaly scan was done at 24 weeks and 6 days age of gestation, revealing, "Pregnancy Uterine 25 weeks and 1 day AOG by fetal biometry, live, singleton breech presentation. Estimated fetal weight = 746 +/- 108 grams. Good cardiac activity (FHR = 141 bpm). Adequate Amniotic fluid volume (SVP 4.4 cm). Anterior placenta, grade II, No previa. No gross congenital anomaly seen."

At 38 weeks and 1 day age of gestation, the patient came in at the emergency room with watery vaginal discharge of 3 hours duration. She was admitted for induction of labor, while closely monitoring the patient's parturition. After 6 hours of labor progression, the patient delivered via normal spontaneous delivery with a final diagnosis of: "Gravida 6 Para 5 (5025) Pregnancy Uterine delivered term cephalic live baby boy, Appropriate for Gestational Age (3600

grams) (APGAR score 8, 9) via Normal Spontaneous delivery with perineal support followed by Intrauterine device insertion; Prelabor rupture of Membranes for nine hours; Grandmultipara; Advanced Maternal age; S/P Exploratory Laparotomy, Salpingectomy Left, for extrauterine - heterotopic pregnancy (June 2022)."



SURGICAL PATHOLOGY CONSULTATION REPORT

Patient Name: [redacted] Accession No.: S-22-2425
Age / Sex:
Case No:
Ward / Institution:
Referring Physician:
Date Received: 27 June 2022
Date Reported: 4 July 2022

Page 1 of 1

PROCEDURE: EXPLORATORY LAPAROTOMY; EVACUATION OF HEMOPERITONEUM; SALPINGECTOMY, LEFT.

HISTOPATHOLOGIC DIAGNOSIS:
- ECTOPIC PREGNANCY, LEFT FALLOPIAN TUBE.

GROSS AND MICROSCOPIC DESCRIPTION
Specimen received in formalin consists of a previously incised, tubular, rubbery tissue measuring 9.0 x 3.0 x 2.5 cm with fimbriae. The serosal surface is tan / dark brown and dull. Sections reveal spongy to rubbery tissues admixed with blood clots. An embryo is noted with a crown-rump length measuring 0.6 cm. No sections were taken from the embryo. Representative sections taken for study, block 1.

Section reveal a dilated tubal wall with attached varisized, avascular chorionic villi and hemorrhagic material. No embryonic tissues seen in the sections examined.

Figure 3: Official Histopathologic Results of the Specimens from the Surgery

The patient's immediate postpartum course was uneventful. The baby was well and breast fed. Both were discharged stable.

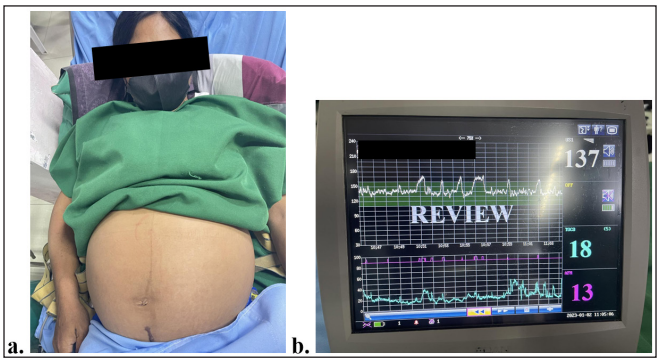


Figure 4: a. Patient GA upon admission. Gravida 6 Para 4 (4024) Pregnancy Uterine at 38 weeks and 1 day Age of Gestation, Cephalic not in Labor, Prelabor Rupture of Membranes for 3 hours; Grandmultigravid, Advanced Maternal Age, s/p Exploratory Laparotomy, Salpingectomy Left for Extrauterine - Heterotopic Pregnancy (June 2022). b. NST reactive.

Case Discussion

Heterotopic pregnancy is defined as a simultaneous development of both intrauterine and extrauterine pregnancy [1]. Diagnosing such cases can be challenging because once an intrauterine pregnancy is seen, a co-existing extrauterine pregnancy may be missed. Or in some cases, a presence of any other sac in the adnexal area with concomitant intrauterine pregnancy may be interpreted as a corpus luteum cyst, which is physiologically occurring, is harmless, and does not require immediate intervention as this fluid-filled mass spontaneously regresses [2]. Diagnosis of a heterotopic pregnancy is inversely proportional with the age of gestation. Most of the time, about 70% of such cases are diagnosed between five and eight weeks age of gestation. The frequency goes down to 20% between 9 and 11 weeks age of gestation, while after 11 weeks age of gestation, frequency is just less than 10% [3].

A high index of suspicion is necessary in order to appropriately diagnose a heterotopic gestation [5]. Most patients with heterotopic pregnancy, along with a positive pregnancy test, present with abdominal pain, peritoneal irritation, vaginal bleeding, an enlarged uterus, and a presence of adnexal mass. However, there are also some diseases that could present as such which can be considered as differential diagnoses. These include miscarriage, a hemorrhagic corpus luteum, adnexal torsion, ovarian hyperstimulation syndrome, an ectopic pregnancy and other non-gynecological conditions like appendicitis, cholecystitis, pancreatitis, or obstructed bowels. Thus, a careful, complete, and thorough history taking and physical exam, along with the aid of laboratories and sonography are imperative in distinguishing a heterotopic pregnancy. Furthermore, some of the risk factors that should be taken into consideration are a prior ectopic pregnancy, pelvic inflammatory disease (PID), abdominal adhesions, history of tubal surgeries, and assisted reproductive therapy (ART) [5]. Other less common risk factors include advanced maternal age, cigarette smoking, endometriosis, presence of intrauterine device (IUD), or an exposure to diethylstilbestrol (DES) in utero [6].

In relation to our case, abdominal pain particularly on the hypogastric area was present together with a positive pregnancy test and peritoneal irritation as noted upon examination. Furthermore, she had no history of ectopic pregnancies as well as pelvic surgeries in the past. Neither a history of pelvic inflammatory disease nor assisted reproductive therapy were noted in the patient.

The initial sonographic findings of the patient pre-operatively revealed an intrauterine pregnancy with an existing right adnexal mass. The laterality was inconsistent to the actual findings intraoperatively, where in the concurrent ectopic gestation was noted on the left. This human error though infrequent can still happen and at times hard to avoid due to technical difficulty. The discrepancy could be due to several factors such as incomplete history taking and physical examination on the patient who probably was not stable nor cooperative enough to accurately disclose all the necessary details about her condition upon initial consult. Technical errors could also lead to such incorrect laterality including the use of a wrong probe and/or transducer, inappropriate setting of the equipment, and the presence of ultrasound artifacts [7]. Nevertheless, the management would still be the same whether the pathological adnexal mass be on the right or on the left. Despite the said paradox, the goal of removing the non-viable extrauterine component while preserving the intrauterine gestation was well accomplished.

The management of heterotopic pregnancy is tailored depending on the patient's hemodynamic status, associated risk factors present, the gestational age at the time of diagnosis, as well as the patient's preference [8]. There are surgical and non-surgical or medical options to choose from. Nevertheless, the ultimate goal is to stabilize the patient by removing the ectopic pregnancy while preserving intrauterine pregnancy and bringing it to viability [5].

Nonsurgical management with methotrexate may be considered to those who are hemodynamically and clinically stable, with minimal abdominal pain, a low level of beta human chorionic gonadotropin (β-hCG) of less than 5,000 IU/L, and

a sonographic finding showing an extrauterine mass measuring less than 3.5 cm with no fetal cardiac activity; provided that the patient is motivated and compliant with post-treatment follow-up. However, such medical regimen could not be applied on this case because it would be detrimental to the existing intrauterine gestation. Methotrexate is a folate antagonist that is contraindicated in cases of deranged ancillary tests such as CBC, renal, and/or hepatic laboratory value, immunodeficiency, active pulmonary disease, peptic ulcer disease, heterotopic pregnancy, hypersensitivity to methotrexate, and/or lactating mothers.

For this reason, the patient on our case was management surgically. The operative approach is indicated in those who are hemodynamically unstable, with an impending or ongoing rupture of the said ectopic gestation, or if nonsurgical management fails. Surgical procedure via laparotomy or laparoscopy could either be salpingostomy if the patient has an unruptured tubal pregnancy and is still desirous of pregnancy in the future; or salpingectomy if the patient has a ruptured ectopic pregnancy with intractable bleeding and a moderately to severely damaged fallopian tube [7]. Seeing a ruptured tubal pregnancy with a damaged fallopian tube intraoperatively, salpingectomy was performed on the patient.

Post-operatively, the dydrogesterone (Oral progesterone) and micronized (vaginal) progesterone given to the patient were beneficial given that she had a history of miscarriage. Recent studies show and recommend giving progesterone to women at a high risk of pregnancy loss or having preterm birth especially during the early first trimester [9,10].

Figure 5: Postpartum. Gravida 6 Para 5 (5025) Pregnancy Uterine delivered term cephalic live baby boy, Appropriate for Gestational Age (3600 grams) (APGAR score 8, 9) via Normal Spontaneous delivery with perineal support followed by Intrauterine device insertion; Prelabor rupture of Membranes for 9 hours; Grandmultipara; Advanced Maternal age; S/P Exploratory Laparotomy, Salpingectomy Left, for extrauterine - heterotopic pregnancy (June 2022)."

Summary

Heterotopic gestation, despite being rare, should be entertained if there are strong evidences for consideration. It must be dealt with utmost attention as proper and timely management is crucial in order to allow a successful intrauterine pregnancy until term without compromising the life of the mother with a simultaneous extrauterine gestation. This paper also emphasizes on the importance of different diagnostic tools such as history taking, physical examination, as well as sonographic and other laboratory tests vital in identification of this uncommon entity. Yes, it is true that extrauterine pregnancy is a usual scenario in our daily practice but unusual to see simultaneous pregnancies at different implantation sites. One like this leading to a viable coexistent intrauterine gestation is definitely one for the record.

In conclusion, there is possibility to have a natural conception in patients with heterotopic gestation who underwent exploratory laparotomy for ruptured extrauterine pregnancy, and at the same time, carry the intrauterine fetus to term with a favorable obstetrical outcome.

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