

Ruptured spontaneous heterotopic pregnancy

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ABSTRACT

تقدر نسبة حدوث حمل داخل و خارج الرحم بمعدل حمل واحد من كل 30,000 حالة حمل تلقائي، وهذه النسبة ترتفع إلى 1% عند حمل واحد في كل 900 حالة عند استعمال عقار كلميفين واستعمال منشطات التبويض مع الوسائل المساعدة على الإنجاب مثل أطفال الأنابيب. يوجد هناك رابط قوي بين حدوث حالات الحمل وحالات العقم. كما أن العوامل التي تساعد على حدوث حالات الحمل خارج الرحم هي حدوث حمل خارج الرحم في السابق، وجود التصاقات في الحوض نتيجة حدوث التهابات سابقة، أو إجراء عمليات جراحية، واستخدام وسائل المساعدة على الإنجاب. المريضة المذكورة في هذا المقال تم تشخيصها في بداية الأمر على أنها حامل بجنين واحد داخل الرحم وتعاني من التهاب المسالك البولية، ولكن المريضة حضرت إلى المستشفى وهي تعاني من آلم حادة لا تتناسب مع التشخيص السابق، و عليه تقرر إجراء أشعة صوتية طارئة، وتبين وجود حمل داخل الرحم مع وجود حمل خارج الرحم أيضاً، ومن ثم تم عمل منظار للمريضة، واستئصال قناة فالوب. في الأيام التالية للمنظار تم عمل أشعة صوتية كما تم عمل تحليل لمعرفة نسبة هرمون الحمل حيث تبين وجود جنين حي داخل الرحم. انتهت الحمل بوفاة الجنين داخل الرحم في الأسبوع السادس عشر، ولذلك تم إجهاض الحمل باستخدام العقاقير الطبية. المواضيع التي تم مناقشتها في هذه المقالة هي ندرة هذه الحالات، والتأخر في التشخيص، أو التشخيص الخطأ لهذه الحالات والعواقب المترتبة على ذلك.

The coexistence of intrauterine and ectopic pregnancy (heterotopic pregnancy) occurs in 1/30,000 of spontaneous pregnancies, 1/900 in Clomiphene citrate induced pregnancies and rises to 1% in assisted reproduction. It is a life-threatening condition with diagnostic and therapeutic complexities. There is strong association between infertility and ectopic pregnancy. Risk factors for ectopic pregnancy are past history, assisted reproduction, and adhesions due to pelvic infection or surgery. Our patient was diagnosed initially as having an intrauterine singleton pregnancy, with urinary tract infection. At presentation pain out of proportion to primary diagnosis led to urgent ultrasonographic review that diagnosed heterotopic pregnancy. It was followed by laparoscopic salpingectomy. Human chorionic gonadotrophin (HCG) after laparoscopy in the subsequent days along with a sonographic evaluation

revealed a viable intrauterine pregnancy. This ended in missed miscarriage and medical expulsion at 16 weeks of gestation. Issues discussed here are rarity, delayed, or misdiagnosis with its sequel.

Saudi Med J 2010; Vol. 31 (4): 445-447

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Received 11th January 2010. Accepted 24th February 2010.

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Heterotopic pregnancy is defined as co existence of intrauterine and ectopic pregnancy. In natural cycles it is a very rare event (1:30,000). It is a life-threatening condition with diagnostic and therapeutic complexities.¹ The first case was reported in 1708 as an autopsy finding.² Here, we report a case of heterotopic pregnancy in a natural cycle, which was misdiagnosed as a normal intrauterine pregnancy. Diagnostic complexity was depicted by some studies like spontaneous triplet heterotopic pregnancy,³ pseudosac with false positive ectopic and hemorrhagic corpus luteum leading to delayed diagnosis.⁴ In our case it was minimal fluid in the pouch of Douglas with heterotopic pregnancy. Our objective in presenting this particular case is to highlight the significance of early detection and management of heterotopic pregnancy to reduce morbidity and mortality associated with it.

Case Report. A 34-year-old lady, para 1+1 presented through the emergency department at King Khalid University & Hospital, Riyadh, Saudi Arabia at 6 weeks of spontaneous pregnancy with dull, lower abdominal pain, burning micturition, urgency, and pyrexia for one day. She had a positive pregnancy test along with an ultrasound from an outside clinic confirming intact intrauterine pregnancy. She was diagnosed by that

clinic as also having urinary tract infection and treated with antibiotics. The previous child was a spontaneous vaginal delivery 10 months ago, and she was on oral contraceptive pills until one and a half months before her presentation. She denies history of smoking, pelvic inflammatory disease, or any previous gynecological surgery. Her pulse was 94 beats per minute and blood pressure 110/70 mm Hg with normal temperature and respiratory rate. At presentation she had soft, non-tender abdomen, and positive bowel sounds. Since the pain was out of proportion to the clinical diagnosis she had an urgent trans-vaginal ultrasound, which showed bilateral corpus luteal cysts, an intrauterine pregnancy and one left sided tubal ectopic pregnancy both with positive fetal heart. There was minimal fluid in the pouch of Douglas (**Figure 1**). Her hemoglobin was 11.1 along with normal coagulation profile. The serum beta human chorionic gonadotrophin (HCG) was 75323 IU/l and mid stream urine was positive for red blood cells, white blood cells 188, protein + 1, and ketones + 1, but nil nitrates. She was on antibiotics at presentation. She proceeded for laparoscopy, which showed 400 ml of intra-peritoneal blood and ruptured ectopic at the left ampullary region. The right tube and ovary were found normal. Left salpingectomy was carried out and the specimen was sent for histopathology. Postoperative recovery was uneventful; a single viable intrauterine pregnancy on ultrasound was confirmed. She was informed at all stages pre and post operatively along with regular follow up advice for antenatal care. Histopathology confirmed fetal tissue in the left tube (**Figure 2**). On follow up, beta HCG was rising and intrauterine pregnancy corresponded to dates at 14 weeks scan. At 16 weeks she presented with per-vaginal spotting, ultrasound confirmed a missed miscarriage; she opted for medical termination of pregnancy. She expelled products of the conception the next day, which

were sent for histopathology confirming products of conception.

Discussion. On literature review we found one case report showing heterotopic pregnancy on clomiphene induction in a 22-year-old woman, leading to rupture and laparotomy due to diagnostic delay, however, later intrauterine pregnancy continued and she delivered at 37 weeks by cesarean section.¹ Our case of spontaneous heterotopic pregnancy was picked up before rupture but ended in missed miscarriage. In another case report⁵ of spontaneous heterotopic pregnancy that was scanned in early pregnancy because of past history of ectopic pregnancy, an intrauterine gestational sac was seen and reassured, but later the patient presented with ruptured ectopic pregnancy that was managed laparoscopically. All these cases emphasize the need for a high index of suspicion in cases with previous ectopic, free fluid and intrauterine sac, even in the absence of adnexal mass. Spontaneous ectopics are missed and delayed in diagnosis due to rarity, which effects prognosis as well. This fact was shown in one retrospective study⁶ that was conducted in the Seoul National University Hospital, Seoul, Korea between 1990-2004 for outcome of intrauterine gestation following surgical treatment for tubal heterotopic pregnancy. They reviewed 30 case reports of tubal heterotopic pregnancy (spontaneous versus assisted reproduction), and a better outcome was depicted in the assisted reproduction group, by higher preoperative systolic blood pressure and live birth, with fewer tubal ruptures. The presence of an intrauterine pregnancy on ultrasound is one of the main causes of misdiagnosis of ectopic, as the incidence of spontaneous heterotopic pregnancy is very rare and the preoperative heterotopic diagnosis is a challenge. History with clinical evaluation can guide to rule out high-risk patients. False-negative results after high-resolution



Figure 1 - Left sided ectopic pregnancy (arrow) on transvaginal scan.

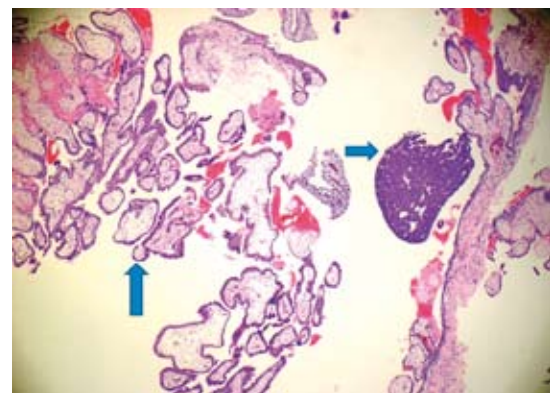


Figure 2 - Histological view of left sided ectopic. Chorionic villi arrow (on left) and immature fetal hepatic, arrow (on right). Hematoxylin & Eosin x 40

transvaginal techniques have been reported due to the presence of ovarian hyper stimulation syndrome masking the ectopic pregnancy in heterotopic pregnancy cases.⁷ Another case report,⁸ detailed the surgical management (salpingotomy via laparotomy) of ruptured, spontaneous heterotopic pregnancy at 11 weeks in a primigravida with no known risk factor, with the outcome being intrauterine pregnancy delivered at 40 weeks. In a case report by Ludwig,⁹ the heterotopic pregnancy was not diagnosed before surgery, she had laparoscopic salpingectomy for ectopic pregnancy with history of previous ectopic of the same tube. On follow up, there was a rising titre of HCG and on ultrasound evaluation a viable intrauterine pregnancy was seen, which resulted in a healthy term baby.⁹

It is well known that both ovarian hyper-stimulation syndrome and heterotopic pregnancy may occur after assisted reproductive techniques. The ectopic pregnancy diagnosis in the presence of an intrauterine pregnancy and ovarian hyper-stimulation syndrome is difficult as symptoms of ectopic pregnancy may be attributed to ovarian hyper-stimulation syndrome, and therefore, ultrasound may not be a sufficiently reliable tool. The difficulty of an accurate clinical and sonographic diagnosis in a symptomatic patient can justify laparoscopy.¹⁰ The treatment option for heterotopic pregnancy is surgery as the first choice to remove the ectopic pregnancy, while non-surgical management includes injection of potassium chloride in the ectopic gestational sac.

In conclusion, in the current era with the increasing use of assisted reproductive techniques and the advent of sonographic facilities, high suspicion and early detection is the primary goal towards successful outcome. Therefore in heterotopic pregnancy, early detection and management can help avoiding fetomaternal compromise with the advent of retaining future fertility potential. It is important to recognize the need to identify other high risk factors with early pregnancy assessment. Documentation and informed consent are of great importance at the same time.

Acknowledgments. We would like to thank Dr. S. A. Zaidi, MD, Registrar, Ultrasound Department, King Khalid University and Hospital, Riyadh, KSA for reviewing the ultrasonographic figures.

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Ethical Consent

All manuscripts reporting the results of experimental investigations involving human subjects should include a statement confirming that informed consent was obtained from each subject or subject's guardian, after receiving approval of the experimental protocol by a local human ethics committee, or institutional review board. When reporting experiments on animals, authors should indicate whether the institutional and national guide for the care and use of laboratory animals was followed.