

Ruptured heterotopic pregnancy with successful obstetrical outcome: A case report and review of the literature

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Summary

Emergency medicine has encountered in the last decades a gradual increase in cases of heterotopic pregnancy (HP) with rupture of the ectopic part. The rise of this entity is mainly due to ovulation induction performed in women undergoing assisted reproductive techniques (ART), but in natural cycles is still rare and unexpected. Diagnosis is often delayed especially in cases where no predisposing factors exist, causing life threatening situations. We report a case of a ruptured heterotopic pregnancy presenting at seven weeks of gestation that was treated with immediate laparotomy. The rest of the pregnancy course was uneventful with spontaneous vaginal delivery of a healthy infant at 39 weeks of gestation. A review of the diagnosis and management of heterotopic pregnancy is also given.

Key words: Heterotopic pregnancy; Natural cycle; Laparotomy.

Introduction

The term heterotopic pregnancy is intended to mean the uncommon condition of simultaneous intrauterine and extrauterine implantation of two different fertilized oocytes. This event is rare in women with natural conception cycles occurring in approximately 1:30,000 pregnancies [1, 2]. A gradual increase has been reported from 1:27,500 cases in 1965 [3], up to 1:3,889 in 1986 [4] reaching an incidence of about 1:100-1:500 [1, 5, 6] in women who conceived with assisted reproductive techniques. Herein, we report a case of a heterotopic right tubal pregnancy in a natural cycle which was first misdiagnosed as a threatened miscarriage.

Case report

A 28-year-old woman, para 0, gravida 1, was referred to our hospital because of lower abdominal pain that deteriorated the last six hours and vaginal bleeding. The patient presented at the emergency room in stable condition and in week 7 of gestation according to her last menstrual period. An abdominal ultrasound performed elsewhere 24 hours before, showed a viable intrauterine fetus with a small amount of fluid in the pouch of Douglas and the referral note reported a probable threatened miscarriage. The medical history was unremarkable. The conception was spontaneous in a natural cycle and there were no risk factors predisposing for extrauterine pregnancy. The urine HCG test was positive. Abdominal examination revealed guarding and rebound in the right quadrant and tenderness in the left abdomen. Bimanual examination disclosed a tender enlarged uterus, with severe pain during cervical movements. Vaginal ultrasound showed a viable fetus with crown-rump-length corresponding to 6th weeks of gestation, the right adnexa with a mass (Figure 1) and the pouch of Douglas filled with echogenic

fluid (Figure 2). The heart rate and blood pressure were still stable. Under general anesthesia an emergency laparotomy by Pfannenstiel incision was performed. An ectopic pregnancy in the proximal right tube was detected and excised with gentle surgical technique. The blood and free clots filling the peritoneal cavity were removed fastidiously. The postoperative course was uneventful and a scan performed 48 hours later disclosed a normal heart pattern of the remaining embryo (Figure 3). The rest of the gestation proceeded without complications. The patient delivered vaginally at 39th weeks after spontaneous labor producing a female infant weighing 3,150 g with Apgar scores of 9,10 and 10 at 1, 5 and 10 minutes, respectively, after birth. The pediatrician followed up the baby for six months and reported normal mental and physical growth.

Discussion

Ectopic pregnancy (EP) is considered a major health problem in women of reproductive age. Before the 19th century, EP was thought to be universally fatal and still now in the USA is the leading cause of maternal death in the first trimester of pregnancy [7]. The factors commonly associated with EP are prior ectopic pregnancy, sexually transmitted disease with pelvic infection, previous pelvic surgery, uterine anomalies, increasing age, cigarette smoking, history of infertility and the use of assisted reproductive technology (ART) [1, 7].

Heterotopic pregnancy, defined as the unusual combination of intrauterine and extrauterine gestation is increased in patients undergoing ART. Lemus, reported a total 70-fold increase, whereas other authors have suggested that the increased number of embryo transfers increases the risk of HP [7]. Dor *et al.* [8] estimated the rate of HP as 1:45 with transfer of more than five embryos. Tummon *et al.* [9] calculated that the odds ratio for the development of HP versus a single EP is increased

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Fig. 1

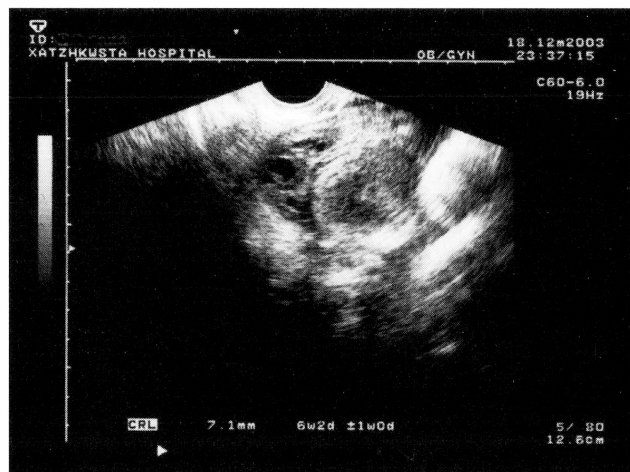


Fig. 2

Fig. 3

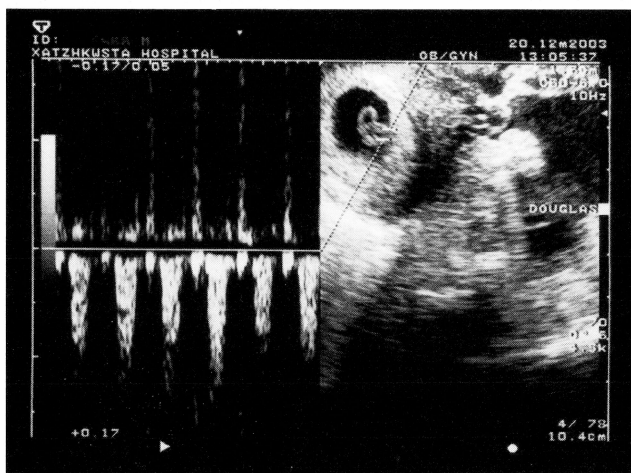


Figure 1. — Transvaginal ultrasound showing an intrauterine pregnancy and the pouch of Douglas filled with echogenic fluid.

Figure 2. — An adnexal mass located at the right side of the uterus with features of an ectopic gestation. This image raised the question of a possible heterotopic pregnancy.

Figure 3. — Transvaginal examination two days after surgery showing an intrauterine embryo with a normal heart pattern.

10-fold when more than four embryos are transferred. In contrast, in spontaneous conception cycles HP is rare with only a few cases mentioned in the literature [10-13]. The fallopian tube is the usual ectopic implantation site [14], although it can be corneal, ovarian, cervical or abdominal [1, 15]. Full-term pregnancy of twins born alive, with one embryo growing inside the uterus and the other one abdominally has also been reported [16, 17].

Approximately 70% of heterotopic pregnancies are diagnosed between five and eight weeks of gestation and the rest of the cases usually up to 11 or 12 weeks [18]. The clinical appearance of HP varies according to the developmental stage of the ectopic embryo. Cases of HP in a spontaneous cycle misdiagnosed as EP at first have been reported due to the non specific symptoms and signs [12]. Often, the only clinical sign is low abdominal pain with mild cramping, which is also the main cause of admittance to hospital for many medical reasons. Final diagnosis, like in our case, is usually established only by exploratory laparotomy [19, 20]. Abdominal pain is present in 83% of heterotopic pregnancies and hypovolemic shock with abdominal tenderness in 13% [18], while other reviews state that HP presenting with acute abdomen and shock is rare [4, 21]. Reece *et al.* [22] defined four presenting signs and symptoms: abdominal pain, adnexal mass, peritoneal irritation and enlarged

uterus. In our report, peritonism was present due to internal bleeding but the heterotopic gestation was detected only after laparotomy.

The trophoblast begins to produce human chorionic gonadotropin (HCG) eight days after conception. The role of biochemical markers like HCG and progesterone is helpful in the diagnosis of EP but not in HP, since there is official placental production from the normal pregnancy covering the production from the ectopic one. The postoperative increase in HCG and progesterone establishes the new diagnosis, thus is not considered excessive measuring of HCG after salpingectomy especially in patients who conceived using ART [13]. This laboratory test also helps avoid dilatation and curettage which is occasionally considered helpful in the diagnosis of EP, but could also terminate a normal intrauterine pregnancy [12].

Often, the intrauterine embryo is detected before the extrauterine one, although a case was reported where the heart activity of the normal embryo was observed one week after the detection of the ectopic one [23]. The use of high resolution, high frequency endovaginal ultrasound is important in the differential diagnosis of pregnant women with acute abdominal pain [12]. In using transvaginal sonography (TVS), it is important to follow a strict technique starting with the visualization of the cervix, endometrial cavity, lateral horns and then passing

to the adnexa and the pouch of Douglas. Concerning intrauterine pregnancy, it is important to distinguish between a true gestational chamber with well defined margins and eccentric location of the decidua from a gestational pseudosac. Unfortunately, the discovery of a gestational sac inside the uterus does not exclude HP [1]. However, ultrasound findings do not have absolute diagnostic value especially in early pregnancy where it is difficult to differentiate a hemorrhagic corpus luteum from a gestational sac located in the fallopian tube. In cases of HP, the thick ring of the tubal wall presents different echogenicity from the ovarian tissue around the corpus luteum [15]. In general, it should be noted that the sonographic evaluation for detection of an ectopic part is associated with false positive and negative results, but its accuracy also depends on the experience of the examiner [2, 24]. Palpating the abdomen in concert with the movements of the vaginal probe seems to increase diagnostic accuracy reaching the examined structures more easily. Most helpful but extremely rare, is the sonographic detection of embryonic heart activity in both intrauterine and extrauterine gestation [13].

In specific cases where ultrasound findings are equivocal careful evaluation could be accomplished by the use of color/power Doppler. By keeping the power levels low and reducing the examination time, the potential injury to the embryo is minimized. In early pregnancy almost 85% of ectopic pregnancies are homolateral to the corpus luteum, thus power Doppler could be useful in distinguishing the typical low resistance high velocity vascularization (tubal ring) from the scarcity of the blood flow at the periphery of the mass and the possible presence of heart activity in the ectopic tubal pregnancy. Overall, it has been reported that color Doppler can increase the accuracy of TVS from 54-71% up to 87-94% [25, 26].

Magnetic resonance imaging (MRI) was evaluated for the detection of EP [27]. MRI allows specific diagnosis of a tubal pregnancy but is not any more useful than ultrasound with color mapping. Furthermore, MRI is one of the most expensive imaging methods, so its use in the diagnosis of EP is contemplated to be excessive [28].

The search of fluid in the pouch of Douglas is presumed clinically important. Puncture of the cul-de-sac is still used to diagnose internal bleeding, although TVS is an efficacious method for blood visualization. Evaluation of the amount (rapid increase or large quantity) of fluid in the abdomen with the characteristic echogenicity is evident of hemoperitoneum [13]. However, if negative findings are present a diagnosis of HP must not be excluded. In our case, we did not perform culdocentesis because of the deterioration of clinical signs, and the presence of fluid in the pouch of Douglas and in the space of Morrison.

There is no agreement about the optimal management of a heterotopic pregnancy. Particular techniques have been reported depending on the location of the extrauterine embryonic sac and the time of diagnosis onset. Rare locations such as cervical or interstitial could be treated conservatively by sonographically guided aspiration or

infusion of potassium chloride, while methotrexate should not be used due to its teratogenic potential [11, 29, 30]. In general, surgical excision by the laparoscopic approach or by laparotomy is the treatment of choice. Lo *et al.* [31] studied 614 patients with tubal pregnancies and concluded that operative time, complications and hospital stay were less for the laparoscopy group, while other authors mention higher complication rates in the same group [32, 33]. In a case of a HP treated by laparoscopic resection of the tubal pregnancy, growth retardation of the surviving embryo was observed associated with the altered intraperitoneal CO₂ environment [34]. In another report the author proceeded immediately to laparotomy after the diagnosis was confirmed laparoscopically [35]. In our case, the pregnant woman was treated with immediate laparotomy due to the worsening of abdominal pain and the presence of internal hemorrhage. The operating time was approximately 30 minutes and the patient did not require further treatment.

In conclusion, heterotopic pregnancy although more frequent remains a diagnostic challenge based on laboratory and imaging methods but mainly on clinical experience. The diagnosis of HP, easily hampered by a false sense of security coming from visualization of the intrauterine pregnancy, is usually established in the operating room. In a recent review the survival rate of intrauterine growing fetuses was reported up to 66% [18], thus it is considered worthwhile to add this rare situation to the differential diagnosis of abdominal pain during pregnancy especially in patients undergoing ART, but also in women with natural conception cycles. Even though miscarriage is sometimes unavoidable, prompt diagnosis could avert life threatening situations and contribute to a favorable obstetrical outcome.

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