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## Case report

## Spontaneous heterotopic triplet pregnancy

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## ARTICLE INFO

## Article history:

Received 22 March 2016

Received in revised form

1 April 2016

Accepted 21 June 2016

Available online 25 August 2016

## Keywords:

Spontaneous pregnancy

Heterotopic ectopic

## ABSTRACT

**Introduction:** Heterotopic pregnancy is a rare condition characterized by the occurrence of two or more simultaneous pregnancies in two or more implantation sites.

**Aim:** To report a case of spontaneous ectopic pregnancy co-existing with twin living intra-uterine pregnancies of 10 weeks gestation.

**Case study:** A 29-years-old woman 10-weeks pregnant presented to the emergency department with acute abdomen. Transvaginal ultrasound revealed di-amniotic intrauterine living twins and right adnexal mass inseparable from the ovary. Laparoscopy revealed intact right tubal pregnancy managed by salpingostomy.

**Results and discussion:** Diagnosis of heterotopic ectopic is difficult as we cannot depend on  $\beta$ -subunit of human chorionic gonadotropin level because of the presence of a co-existent intrauterine pregnancy which affects the hormonal level and the false sense of security that may be present on visualizing an intrauterine pregnancy by ultrasonography (USG), therefore, it is important to visualize the adnexa even in the presence of an intrauterine pregnancy to avoid missing a possible co-existent ectopic pregnancy. Surgery is the gold standard treatment. Other treatment modalities as laparoscopic or transvaginal USG guided injection of hyperosmolar glucose or potassium chloride have less success rates.

**Conclusions:** It is important to scan the adnexa even in the presence of an intrauterine pregnancy to avoid missing a possible co-existent ectopic pregnancy. Surgery is the gold standard treatment, however, other modalities were described with less success.

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## 1. Introduction

Heterotopic pregnancy is a rare condition characterized by the occurrence of two or more simultaneous pregnancies in two or more implantation sites.<sup>1,2</sup> Heterotopic pregnancy has a prevalence of 0.6–2.5 cases in 10 000 pregnancies,<sup>3</sup> with

increased incidence in women taking ovulation induction medications, and a greater incidence in women undergoing assisted reproduction techniques, e.g., in vitro fertilization, reaching up to three cases in 100 pregnancies. Transfer of more than 4 embryos raises the risk to 1 case in 45 pregnancies. In natural conceptions, the incidence is a lot lower reaching 1 case in 30 000 pregnancies.<sup>4</sup>

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## 2. Aim

To report a case of spontaneous ectopic pregnancy co-existing with twin living intra-uterine pregnancies of 10 weeks gestation.

## 3. Case study

A 29-years-old woman married for a year, 4th gravida with previous 3 successive miscarriages (all first trimester miscarriages) with spontaneous 10 weeks pregnancy presented to the emergency department in As-Salam International Hospital with acute abdominal pain and tenderness in the right iliac fossa. Her blood pressure was 100/60 mmHg and pulse was 102 beats per minute. She was initially diagnosed as acute appendicitis, blood was taken for full-blood count. Pelvi-abdominal ultrasonography (USG) examination showed a mass adjacent to the right ovary, so transvaginal USG was done. It showed di-amniotic living twins crown rump length equivalent to 9 weeks and 3 days (Fig. 1), two unilocular simple ovarian cysts – one on the right measuring about 2 cm and the other on the left measuring about 4 cm – in addition to, right adnexal mass inseparable from the ovary measuring about 8 × 4 cm (Fig. 2). Full blood count revealed hemoglobin of 10.7 g/dL, white cell count was 5 600/mm<sup>3</sup> and platelet count was 227 000/mm<sup>3</sup>.

Differential diagnosis of this case included acute appendicitis (phlegmon) and the rare possibility of heterotopic pregnancy. In view of the above findings the decision was taken to perform a diagnostic laparoscopy with involvement of both consultant gynecologist and consultant surgeon, which, revealed intact right tubal pregnancy (Fig. 3) with peri-fimbrial blood clot and moderate amount of blood in the pelvis, in addition to bilateral simple ovarian cysts.

Laparoscopic salpingostomy was done as this was the patient's preference. Both ovarian cysts were left as they were thought to be corpus luteum of pregnancy.



Fig. 1 – Diamniotic twin intrauterine pregnancy.

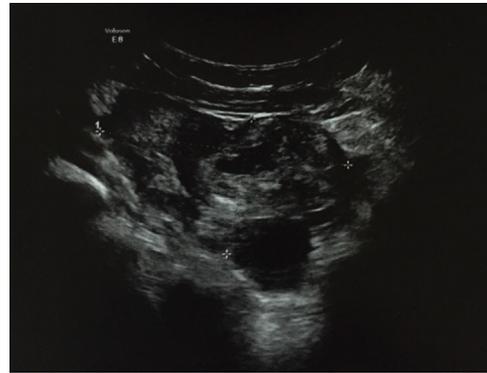


Fig. 2 – Right adnexal mass and unilocular ovarian cyst.



Fig. 3 – Right tubal pregnancy.

Postoperative USG examination was done after 2 days which confirmed that both intrauterine pregnancies were still viable and no free fluid in the abdominal cavity so the drains were removed. Postoperative hemoglobin dropped to 9.5 g/dL. Recovery was uneventful. Two weeks after operation another scan was done, which revealed that both twins are viable. The pregnancy continued until 34 weeks. She delivered two healthy female babies following a spontaneous vaginal delivery.

## 4. Results and discussion

Spontaneous heterotopic pregnancy is very rare with an incidence of 1 in 30 000 pregnancies.<sup>4</sup> Risk factors are previous ectopic pregnancy, cigarette smoking, conditions causing tubal damage as endometriosis, tubal surgery and pelvic inflammatory disease, and assisted reproductive techniques,<sup>5-8</sup> the latter cause being responsible for the rise of the risk of ectopic pregnancy reaching up to one percent in this subgroup.<sup>9,10</sup> Spontaneous heterotopic triplets pregnancy with twin intrauterine pregnancies and a third tubal ectopic pregnancy are very rare.<sup>9,11</sup>

Diagnosis of heterotopic pregnancy can be both difficult and deceiving. We cannot depend on  $\beta$ -subunit human chorionic gonadotropin level because of the presence of a

co-existent intrauterine pregnancy which affects the hormonal level,<sup>9,12</sup> and the false sense of security that may be present on visualizing an intrauterine pregnancy by USG, therefore, it is important to visualize the adnexa even in the presence of an intrauterine pregnancy to avoid missing a possible co-existent ectopic pregnancy.<sup>1,10</sup> Li et al.,<sup>13</sup> found 100% specificity and 92.4% sensitivity for the detection of heterotopic pregnancy in their cohort, however, other studies had lower detection rates ranging between 41% and 84% due to the similarity in appearance between an anembryonic sac and a corpus luteum cyst.<sup>8,10,14,15</sup>

Surgery is the gold standard in the treatment of heterotopic pregnancies either via laparoscopy or laparotomy on case by case basis.<sup>1–3</sup> However, other treatment modalities were described as laparoscopic or transvaginal USG guided injection of hyperosmolar glucose or potassium chloride with less success rates raising concerns about those modalities.<sup>16</sup> In the current case we chose the laparoscopic approach being associated with faster recovery, less risk of uterine manipulations, good visualization, absence of severe hemoperitoneum, and above all the patient was not shocked. Salpingostomy was done according to the patient's choice.

## 5. Conclusions

Spontaneous heterotopic pregnancy is rare. It is important to scan the adnexa even in the presence of an intrauterine pregnancy to avoid missing a possible co-existent ectopic pregnancy. Surgery is the gold standard treatment, however, other modalities were described with less success.

## Conflict of interest

None declared.

## Acknowledgments

Authors declare that they neither have a conflict of interest nor received any financial support.

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