

# Term, live, primary ovarian pregnancy: A case report

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## ABSTRACT

Primary ovarian pregnancy accounts for less than 1% of all cases of ectopic pregnancies. Its diagnosis is most commonly made intra-operatively and the approach in its management has been geared towards conservative measures such as oophorectomy and resection. Use of intrauterine device still remains the most established risk factor for the development of ovarian pregnancy. The diagnosis is established following the criteria first described by Spiegelberg in 1878. Several cases of ovarian gestation have been described in literature, although very few cases with live term fetus have been reported. Majority of the cases were diagnosed intra-operatively, and the management was tailored depending on the complexity of each of the different cases. This paper reports a case of primary ovarian pregnancy with a live term fetus, which was only diagnosed intra-operatively. Total hysterectomy with right salpingo-oophorectomy was performed due to difficulties encountered brought about by dense adhesions.

*Keywords: Ectopic pregnancy, ovarian pregnancy, primary ovarian pregnancy*

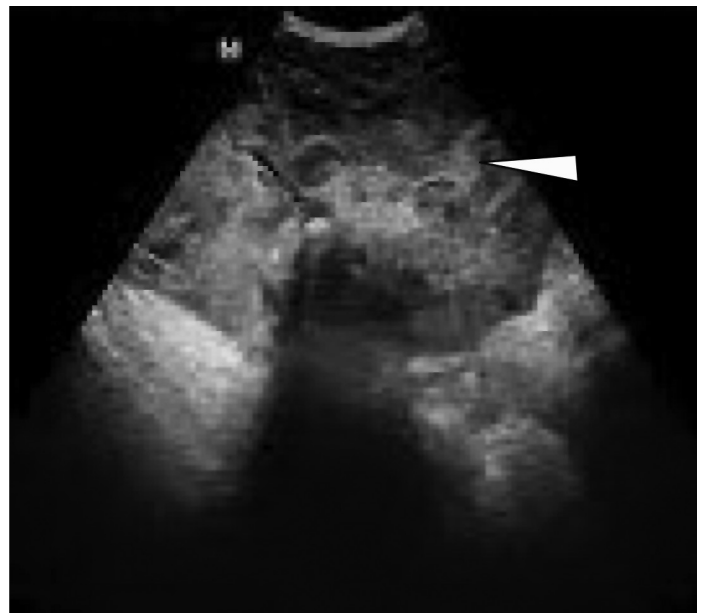
## INTRODUCTION

Ovarian pregnancy accounts for 0.5 to 1% of all cases of ectopic gestation.<sup>1</sup> Majority of the cases result in rupture, with 65% of cases rupturing by 8 weeks age of gestation, and 91% by 13 weeks.<sup>2</sup> Very few cases of ovarian pregnancies reaching term have been reported and only a few number of infants survived. We present a case of primary ovarian pregnancy with a live term fetus, which was only diagnosed intra-operatively.

## CASE REPORT

A 27 year-old gravida 3 para 2 (2002) was seen initially at 18 weeks age of gestation by amenorrhea for her sole prenatal visit at a local health center. Ancillary tests done at that time were limited to a complete blood count and urinalysis, both of which revealed normal results. A biometry was also done, which revealed a single live intrauterine pregnancy in cephalic presentation, 21 weeks and 2 days by biparietal diameter and 18 weeks and 5 days by femoral length, with good cardiac and somatic activities (Figure 1). The placenta was anterofundal, grade I and the amniotic fluid volume was normal. The antenatal course was uneventful.

She was admitted at 36 weeks age of gestation for painful uterine contractions, which started around 16 hours prior to admission. There was report of good fetal movement and there was no history of bloody and watery vaginal discharge. She was received with stable vital signs



**Figure 1.** Ultrasound image showing the absence of a myometrium surrounding the gestation.

and essentially normal systemic physical examination findings except for a hemic murmur on auscultation. Abdominal examination revealed an enlarged, globular abdomen with a fundic height of 25 cm and an estimated fetal weight of 1400 to 1600 grams. Occupying the fundal area was the breech, and the head was noted to be the presenting part. No fetal heart tone was appreciated. On internal examination, the cervix was 1 cm dilated with beginning effacement. The presenting part was not

palpated. The consideration at the time was abruptio placenta, rule out fetal death in utero. The plan was to terminate the pregnancy by abdominal delivery.

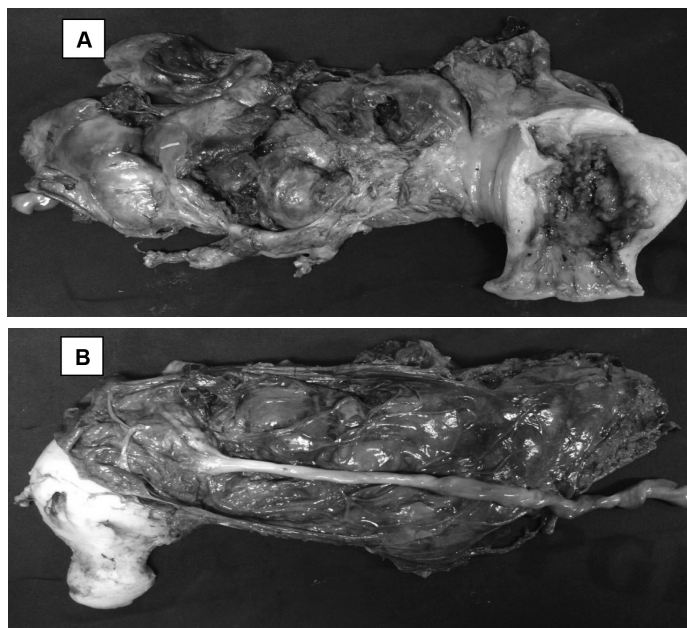
Upon opening the peritoneum, there was sudden gush of hemoperitoneum. The placenta was located directly inferior to the peritoneum. Posterior to the placenta was the amniotic sac which contained clear amniotic fluid, within which was a live baby boy, 37 weeks by pediatric aging, small for gestational age, with an APGAR score of 8 becoming 9. The baby had bilateral cleft lip and palate. The placenta was noted to be continuous with the left fundal area of the ureus, extending up to the area of the right broad ligament and right infundibulopelvic ligament. It was likewise adherent to the appendix and the surrounding small bowels (Figure 2). Careful adhesiolysis was done after which a total hysterectomy with right salpingo-oophorectomy was performed.



**Figure 2.** Photo showing the placenta to be continuous with the left fundal area and adherent to the appendix and the small bowels.

On inspection of the specimen, there seemed to be no normal ovarian tissue attached to the placenta. The fallopian tube was seen on the superior portion of the placenta measuring around 7 x 0.5 cm. The uterus was small with smooth, tan serosal surface measuring 10 x 10 x 8 cm. The cervix measured 4 x 3 x 3 cm. Cut section of the right adnexa showed placenta-like tissues which seemed to invade the right lateral and posterior aspects of the uterine wall (Figure 3). The uterus, right adnexa, and placenta were sent for histopathological examination. The histopathological diagnosis was ovarian pregnancy, a mature singleton placenta, and decidualized endometrium.

The patient suffered from hypovolemic shock and acute kidney injury secondary to renal hypoperfusion following an intraoperative blood loss of 3000 cc. These were all eventually corrected. However, on the fifth post-operative day, she developed a sudden onset of



**Figure 3.** Photo showing the placenta-like tissues which seemed to invade the right lateral and posterior aspects of the uterine wall. There was no intrauterine gestation on cut section. **A.** Anterior view. **B.** Posterior view.

unilateral temporal headache with a pain severity score of 10/10, followed by an episode of seizure described as having blank stares, fluttering eyelid, preferential gaze to the left, and generalized tonic clonic movement lasting for one minute. Post-ictal, she was noted to have weakness on both upper and lower extremities but with no other neurologic deficits. A cranial computed tomography scan revealed an acute hypodensity on the right parieto-temporo-occipital area. The assessment was acute cerebrovascular disease, consider central venous thrombosis with secondary symptomatic seizures. The patient was maintained on prolonged anticoagulation and was eventually discharged on the 18th post-operative day. The patient refused further work-up and management and was eventually lost to follow up.

## DISCUSSION

Ectopic gestation accounts for 10% of the overall maternal mortality. The most common site are the fallopian tubes.<sup>3,4</sup> Primary ovarian pregnancy, though a rare entity, accounts for 0.5 to 1% of all cases of ectopic gestation.<sup>1</sup> This figure was based on diagnosis made using the histologic criteria established by Spielberg in 1878. Recently, it has been suggested that the actual incidence may be higher at up to 1 in 1400 deliveries if the criteria other than those of Spielberg are taken into consideration. These criteria combine biochemical and USG findings and include the following: a) serum  $\beta$ hCG level  $\geq 1000$  IU/L b)

no gestational sac in uterine at transvaginal ultrasound c) ovarian involvement should be confirmed on exploration, with bleeding, visualization of chorionic villi or presence of atypical cyst as the ovary d) normal tubes e) absence of serum  $\beta$ hCG after treatment of ovary.<sup>5</sup>

The distinction between a primary and a secondary ovarian pregnancy is made depending on where the fertilization of the ovum took place. It is considered a primary ovarian pregnancy when the ovum is fertilized while it is still in the follicle, and is a secondary ovarian pregnancy when the fertilization occurs in the fallopian tube and subsequently implants in the ovarian stroma.<sup>6</sup>

Several theories have been suggested regarding the cause of ovarian pregnancy. The most widely discussed is the theory of reflux or reverse migration. This occurs when there is reflux of the conceptus after a normal fertilization from the fallopian tube occurs.<sup>7</sup> This indirectly explains why current use of an intrauterine device was identified as a major risk factor associated with the occurrence of ovarian pregnancy.<sup>7</sup> Studies have shown that an intrauterine device was found in 57-90% of patients with a primary ovarian pregnancy compared to 14-30% of patients with a nonovarian extra uterine pregnancy.<sup>8</sup>

For years, the use of an intrauterine device was the sole risk factor mentioned in literature. Recent literature, however, have noted increased incidence of ovarian pregnancy among patients who have undergone assisted reproductive technologies.<sup>9,10</sup> Other risk factors that have now been implicated in the development of ovarian pregnancy include previous pelvic inflammatory disease, endometriosis and previous abdominal surgeries including a history of tubal ligation.<sup>9-12</sup> None of these risk factors was present in our index case.

Among patients who consult in the first trimester, the most common presenting symptom is acute abdominal pain followed by vaginal bleeding.<sup>10</sup> In some instances, an adnexal mass may be palpable on bimanual examination.<sup>8</sup> Around 75% of cases rupture in the early first trimester particularly at around the 40th day of gestation as a result of the trophoblastic cells eroding into the ovarian microvessels.<sup>9,10</sup> In such cases, they are often misdiagnosed as a ruptured corpus luteum cyst<sup>8,10,13</sup> If the embryo is able to establish blood supply communication with the ovary, then the ovarian pregnancy may continue to the second and third trimester.<sup>14</sup> Massive hemorrhage complicates these pregnancies and most patients present with circulatory collapse.<sup>10,13</sup> In our index case, the placenta seemed to have derived its blood supply from the uterus, ovary and infundibulopelvic ligament thus enabling the pregnancy to proceed until term. It was fortunate that she was still hemodynamically stable at the time of admission despite the presence of hemoperitoneum on exploratory laparotomy.

Ultrasound in the diagnosis of ovarian pregnancy can only be beneficial if the patient comes in unruptured state. The following sonographic criteria have been suggested: a wide echogenic ring with an internal echolucent area on the ovarian surface; the presence of ovarian cortex, including corpus luteum or follicles around the mass; and the echogenicity of the ring usually greater than that of the ovary itself.<sup>10</sup> Unfortunately, most patients present with ruptured ectopic pregnancy with circulatory collapse thus making pre-operative diagnosis difficult. For our index case, a first trimester ultrasound was not done and the second trimester ultrasound totally missed out the diagnosis.

The Spiegelberg criteria (1878) is currently used for the definitive diagnosis of an ovarian pregnancy. The criteria requires presence of the following histologic features: (a) the tube on the ipsilateral side of the pathology has to be entirely normal, (b) the gestational sac has to be situated anatomically within the ovary, (c) both the ovary and the gestational sac have to be connected to the uterus through the utero-ovarian ligament, and lastly (d) placental tissues, on histology, should be identified admixed with the ovarian cortex.<sup>15</sup> Our case fulfilled all four criteria of Spiegelberg hence supporting the diagnosis of an ovarian gestation.

Management for ovarian pregnancy vary depending on the presentation of the patient. Through the years, conservative surgery has been the management of choice. When diagnosed early, cystectomy or wedge resection via laparoscopy or laparotomy are preferred as opposed to oophorectomy. The use of chemotherapeutic agents such as methotrexate or etoposide still remains controversial.<sup>10,13</sup>

Our patient underwent total hysterectomy with unilateral salpingo-oophorectomy owing to the difficulty of the procedure secondary to the adhesions between the placenta and the uterine serosa and surrounding structures. Massive hemorrhage was encountered intraoperatively, since hypertrophied vessels, constrict very poorly in these cases. Arterial embolization of specific bleeding sites has been shown to be life-saving in some cases. Some authors have suggested leaving the placenta behind followed by administration of methotrexate to aid in placental involution.<sup>13</sup> This strategy has yielded varied results.

## SUMMARY

Ovarian pregnancy is a rare form of ectopic gestations. Majority of the reported cases result in early first trimester rupture. Hence, ovarian pregnancy reaching term is a rare occurrence especially if a live fetus is delivered. Pre-operative diagnosis is difficult and most of the cases are still diagnosed intraoperatively. Spiegelberg described

the diagnostic criteria used for such cases in 1878 and his criteria is still used today for the definitive diagnosis of an ovarian pregnancy. Conservative surgical approach if the

management of choice but surgical treatment should still be individualized depending on the patient's presentation and hemodynamic status. ■

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