Thai Journal of Obstetrics and Gynaecology
July 2025, Vol. 33, No. 4, pp. 360-365.

CASE REPORT

Ruptured Primary Ovarian Pregnancy in Rural Setting: A rare case report

Glenardi Glenardi, M.D.*,
Hendri Tanjaya, M.D.*,
Fredrico Patria, M.D.**,
Fitriyadi Kusuma, M.D.***,
Weny Yusnita, M.D.****,

** Department of Medicine, School of Medicine and Health Sciences, Atma Jaya Catholic University of Indonesia, North Jakarta, DKI Jakarta, Indonesia*
*** Department of Obstetrics and Gynecology, Kramat Jati National Police Hospital, East Jakarta, DKI Jakarta, Indonesia*
**** Department of Obstetrics and Gynaecology, Dr. Cipto Mangunkusumo Hospital, Greater Jakarta, DKI Jakarta, Indonesia*
***** Department of Pathological Anatomy, Kramat Jati National Police Hospital, East Jakarta, DKI Jakarta, Indonesia*

ABSTRACT

Primary ovarian pregnancy is a rare type of ectopic pregnancy that has been associated with higher chance of rupture compared to other types. We present a 23-year-old woman with severe lower abdominal pain and 4 weeks amenorrhea. On physical examination, she was clinically unstable with distended abdomen and tenderness on palpation. Bimanual pelvic examination revealed cervical motion tenderness, left adnexal mass, and adnexal tenderness. Human chorionic gonadotropin test was positive, and transabdominal ultrasound showed no gestational sac in uterine cavity with massive fluid collection surrounding the uterus that hinder the identification of the ovaries. The exploratory laparotomy revealed a mass on the left ovary with active bleeding. Wedge resection was conducted, and histopathological examination showed chorionic villi and trophoblastic cells embedded in ovarian stroma, confirming the diagnosis of primary ovarian pregnancy. Differentiating ovarian pregnancy from tubal pregnancy, corpus luteum cyst, or ovarian endometrioma via ultrasound remains challenging.

Keywords: emergency, ectopic pregnancy, ovarian pregnancy, pregnancy complication.

Correspondence to: *Glenardi Glenardi, M.D. Department of Medicine, School of Medicine and Health Sciences, Atma Jaya Catholic University of Indonesia, Pluit Raya No. 2, North Jakarta 14440, Indonesia. Email: glenardihalim@gmail.com*

Received: 30 July 2024, **Revised:** 26 December 2024, **Accepted:** 7 February 2025

Introduction

Primary ovarian pregnancy is extremely rare form of ectopic pregnancy, with an approximately incidence of 0.15 - 3.5% of all ectopic pregnancy and 1 in 7,000 to 40,000 live births[1, 2]. Despite of being one of the rarest forms of ectopic pregnancy, it is reported to have a higher chance of rupture compared to tubal pregnancy, which is the most common form of ectopic pregnancy[3]. Therefore, preoperative diagnosis is crucial in the management of ovarian pregnancy to prevent poor outcomes and severe complications. However, it is challenging as the clinical presentations of ovarian pregnancy are frequently similar to tubal pregnancy, hemorrhagic corpus luteum, complicated ovarian cyst or granulosa cell tumor[4-6].

Hence, it is important to have an ovarian pregnancy as one of the differential diagnoses before proven otherwise. Herein, we present a case of ruptured ovarian pregnancy in a primigravida patient with no pre-existing risk factors.

Case report

A 23-year-old unmarried female was admitted to the hospital with severe abdominal pain and a history of amenorrhea for 29 days. She had associated symptoms of dizziness, nausea and several episodes of vomiting but there was no history of vaginal bleeding, loss of consciousness, urinary or gastrointestinal tract symptoms. The patient reported being sexually active and had not use any contraceptive methods. There was also no history of symptoms suggestive of pelvic inflammatory disease or other medical conditions. The patient admitted that she had never been pregnant before and had not undergone any previous surgery. There was no significant family history. On physical examination, she looked distressed and pale. Her body temperature was 36.5°C, blood pressure was 90/60 mmHg with a pulse rate of 116 beats per minute, and respiratory rate were 24 breaths per minute. The abdomen was distended and tender on palpation with muscular defence and rebound tenderness. The pelvic examination revealed cervical motion tenderness, left adnexal mass, adnexal tenderness and fullness in the pouch of Douglas. The bedside qualitative urine human chorionic gonadotropin (hCG) test was positive. Laboratory analysis revealed a decreased hemoglobin level (9.6 g/dL) and elevated white cell count (27,430 cells/μL). Transabdominal ultrasound showed normal uterus size with no gestational sac in the endometrial lining, massive fluid collection in the pouch of Douglas and surround the uterus and adnexa indicating hemoperitoneum, and no ovaries were able to be identified. Transvaginal ultrasound could not be performed due to the unavailability of the probe in primary care settings. Additionally, the patient's condition was deteriorating, making it impossible to undergo another ultrasound examination.

Based on these findings, patient was suspected of having a ruptured ectopic pregnancy. After securing blood products and obtaining informed consent, an emergency exploratory laparotomy was conducted. An 8 - 10 cm Pfannenstiel incision was made two fingerbreadths above the pubic bone. Approximately 1,200 ml of blood and blood clots was present at the peritoneal cavity which was then aspirated. After lavage, we found an approximately 2x2 cm mass on the left ovary with active bleeding. Both fallopian tubes and right ovary appeared normal (Fig. 1).

Wedge resection was then performed, and the extracted tissue was sent for histopathological analysis. During the operation, the patient received two packed red blood cells. The patient remained hemodynamically stable during the postoperative period and was discharged on the second postoperative day. The histopathological examination revealed a predominantly hemorrhagic area containing chorionic villi and trophoblastic cells embedded in ovarian stroma, which was consistent with the finding of primary ovarian ectopic pregnancy (Fig. 2).



Fig. 1. Intraoperative findings of a ruptured primary ovarian pregnancy; a gestational sac in the left ovary (arrow).

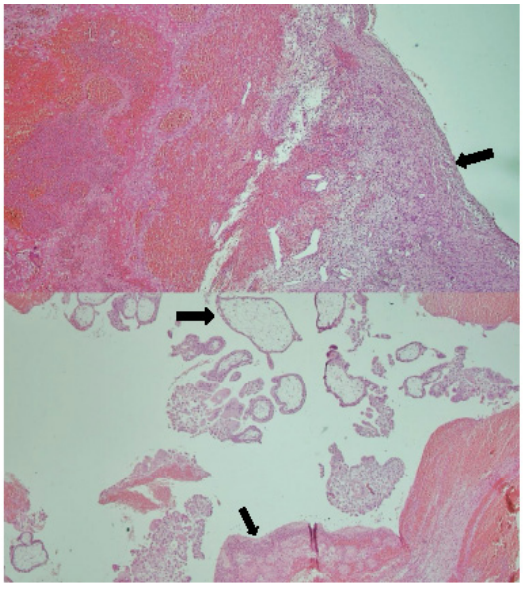


Fig. 2. Histopathology result of the specimen; section shows chorionic villi, and trophoblastic cells are embedded in left ovarian stroma (arrow) [hematoxylin and eosin stain at magnification of 40 X]

Discussion

Ovarian pregnancy is a rare type of ectopic pregnancy which the gestational sac is implanted and developed in ovary. The implantation anomaly is suggested due to the defective follicular extrusion or disruption of ovum transport pathway, which then causes fertilization of the ovum in the peritoneal cavity and implantation on the surface of the ovary[7, 8]. Although the exact cause of ovarian pregnancy is unknown, there are several risk factors that may be associated with this condition, such as use of an intrauterine device, assisted reproductive technology, multipara, history of pelvic inflammatory disease, and history of pelvic or abdominal surgery[9,10]. However, the reported patient did not have these risk factors. This finding was similar to other studies in which most cases of ovarian pregnancy can occur without having any risk factors[10,11]. Despite the low incidence of

ovarian pregnancy, it is among the leading causes of maternal morbidity and mortality in the first trimester[12]. The ovary consists of abundant blood vessels and lacks muscle fibers, which make it easier to rupture and lead to intraabdominal hemorrhage and hypovolemic shock. A ruptured ovarian pregnancy commonly present with sudden severe abdominal pain, dizziness, and syncope, which was the case in the reported patient[3, 6]. Therefore, an early diagnosis and immediate intervention were the key to produce better prognosis.

Preoperative diagnosis is usually done clinically with the supporting data from biochemical and ultrasound findings. Combination of the ectopic pregnancy's classic clinical triad (abdominal pain, vaginal bleeding, and amenorrhea) along with elevated βhCG level, and the absence of gestational sac in the uterus on ultrasound is sufficient to consider ectopic pregnancy as a differential diagnosis[10]. However, it is often challenging to differentiate an ovarian pregnancy from a tubal pregnancy, corpus luteum cyst, or ovarian endometrioma sonographically[4, 6].

Comstock et al proposed an ultrasound diagnostic criteria for ovarian pregnancy, which are the following: a large echogenic ring with an internal echolucent area on the surface of the ovary, the ring has a greater echogenicity than the ovary, and the investigated mass is surrounded by ovarian cortex, including follicles and corpus luteum[13]. Unfortunately, it is almost impossible to diagnose a ruptured ovarian pregnancy preoperatively using ultrasound because the formation of blood clot can hinder identification of the ovaries, which was the case in this patient[6].

The ultrasound examination of this patient was also unable to clearly identify the ovaries, thus the diagnosis had to be made intraoperatively and confirm postoperatively as in most cases of ovarian pregnancies. The Spiegelberg's criteria are used to diagnose ovarian pregnancy, which is a combination of anatomic and histopathological findings. These are the following criteria: the ipsilateral fallopian tube and fimbria must be intact and separate from the ovary, the gestational sac must occupy the normal position of ovary, the ovary and gestational must be attached to uterus through the ovarian ligament, the ovarian tissue must be present in the wall of gestational sac and proven histologically[14]. In this case, the Spigelberg's criteria were met thus the diagnosis of ovarian pregnancy was concluded.

Primary ovarian pregnancy refers to the implantation of a fertilized ovum on the outer surface of the ovary. This type of pregnancy typically ends in rupture during the first three months. Primary ovarian pregnancy typically occurs due to ovulatory dysfunction, where the ovum is fertilized while still inside the follicle before it is expelled from the ovary. In contrast, secondary ovarian ectopic pregnancy refers to the occurrence of fertilization within the fallopian tube, followed by the conceptus being expelled and implanted in the ovarian stroma[15].

Roy et al found that cases of secondary ovarian pregnancy can be established when there is evidence of a previous ectopic pregnancy in the fallopian tube which then regurgitates implantation into the ovary. In this case, they found a tubo-ovarian mass which was then subjected to histopathological examination of the tubo-ovaries and found a trophoblastic tissue in both the ovary and fallopian tube of the location of the ectopic pregnancy. This supports the diagnosis of secondary ovarian pregnancy[16]. In our case, we did not find any abnormality or tubal abortus mass in both fallopian tubes, so histopathological examination of the fallopian tubes was not done, considering the fertility of the patient. In addition, the Spielberg criteria were all met in our case. Hence, a diagnosis of primary ovarian pregnancy was made.

The management of ruptured ovarian pregnancy is done surgically with the primary goal to remove the ectopic pregnancy while preserving the ovary tissue. It can be accomplished by dissection of trophoblast off the ovary or ovarian wedge resection. In the case of excessive bleeding or coexisting ipsilateral ovarian

pathology or a request for removal of the ovary from the patient, an oophorectomy may be performed[10,17]. In this case we performed an emergency laparotomy because the site of the ectopic pregnancy could not be identified preoperatively along with the signs of circulatory collapse indicating the need for urgent management. An ovarian wedge resection was done to preserve the future fertility.

Several case reports have addressed primary ovarian pregnancy, and we present a case identified in a primary care setting that meets the Spielberg criteria for primary ovarian ectopic pregnancy as in the case of Phupong et al, which was also confirmed by histopathology[18]. In the case reported by Meethong et al, histopathological examination was not performed leading to an uncertain diagnosis as the Spielberg criteria were not fulfilled[19]. For the management of ovarian pregnancy, some studies have reported performing laparoscopic surgery[20, 21]. However, these procedures can be challenging due to the risk of significant intraabdominal bleeding, which can obstruct visualization, as demonstrated in this case. Additionally, our patient presented with hemodynamic instability, necessitating urgent intervention[11]. Therefore, laparotomy is still relevant in such emergency cases as in our case and Bharti et al[22].

In the cases of ruptured ovarian pregnancy, urgent management are often required which can lead to more frequent misdiagnosis due to its rarity. This case report accentuates the importance of considering an ovarian pregnancy as a differential diagnosis in patients presenting with classic signs of ectopic pregnancy.

Potential conflicts of interest

The authors declare no conflicts of interest.

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