

## Case Report

## Management of Spontaneous Cornual Heterotopic Pregnancy in Low-Resources Setting

### Tata Laksana Kehamilan Heterotopik Kornual Spontan pada Keadaan Sumber Daya Terbatas

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#### Abstract

**Objective:** To report management of spontaneous cornual heterotopic pregnancy in low-resources setting in Ende District, Flores, East Nusa Tenggara.

**Methods:** Case report.

**Case:** A 34 year old primigravida with history of 8-9 weeks amenorrhea came to Obstetrics ER with chief complaint of vaginal bleeding and lower abdominal pain. Ultrasound shows intrauterine pregnancy (IUP), an ectopic pregnancy (EP) in right uterine cornu, and free fluid in hepatorenal space, splenorenal space, and pouch of douglas suggesting the occurrence of hemoperitoneum and heterotopic pregnancy. We performed cornual resection by laparotomy and administered progesterone orally before and after the surgery. Successful outcome was achieved.

**Discussion:** Heterotopic pregnancy (HP) rarely occurs, especially in natural conception. Thus, early diagnosis and treatment of HP are quite a challenge for physicians especially in rural area. Due to the condition of our patient and limited resources, laparotomy was conducted to remove the EP, rather than laparoscopy despite its advantage to lower risk of IUP abortion. Progesterone was then administered orally to prevent threatened abortion of the IUP.

**Conclusion:** Despite its challenge in diagnosing and treating HP, it is a life-threatening condition that requires accurate and prompt treatment. The treatment goal is to remove the EP and preserve the IUP. Treatment of choice should be decided by taking the patient's condition and availability of resources into account. Surgical along with administration of progesterone before and after the surgery would likely improve the outcome of the patient and the intrauterine pregnancy.

**Keywords:** cornual resection, heterotopic pregnancy, laparotomy, low-resources setting, progesterone.

#### Abstrak

**Tujuan:** Untuk membahas tentang penatalaksanaan kehamilan heterotopik kornu spontan di daerah dengan sumber daya rendah khususnya di Kabupaten Ende, Flores, Nusa Tenggara Timur.

**Metode:** Laporan Kasus

**Kasus:** Seorang perempuan primigravida usia 34 tahun dengan riwayat amenore minggu ke-8 dan 9 datang ke IGD Obygn dengan perdarahan pervaginam dan nyeri perut bagian bawah. Temuan USG menunjukkan kehamilan intrauterin (KIU), kehamilan ektopik (KE) di tanduk rahim kanan, dan cairan bebas di ruang hepato-renal, spleno-renal, dan cavum douglas. Hal ini menunjukkan terjadinya hemoperitoneum dan kehamilan heterotopik. Reseksi kornu dengan laparotomi dilakukan dan pasien diberikan progesteron secara oral sebelum dan setelah operasi. Luaran baik berhasil dicapai.

**Diskusi:** Kehamilan heterotopik jarang terjadi, terutama pada konsepsi alami. Sehingga diagnosis dan tata laksana KH sejak dini menjadi tantangan bagi para dokter, terutama di daerah terpencil Karena kondisi pasien dan sumber daya, laparotomi dilakukan untuk mengangkat KE, daripada laparoskopi meskipun keuntungannya dalam menurunkan risiko keguguran KIU. Progesteron kemudian diberikan secara oral untuk mencegah terjadinya keguguran terancam dari KIU.

**Kesimpulan:** Terlepas dari tantangan untuk diagnosis dan tatalaksananya, KH adalah kondisi yang mengancam jiwa yang membutuhkan penanganan yang akurat dan segera. Tujuan tatalaksananya adalah untuk mengangkat KE dan mempertahankan KIU. Pilihan tata laksana harus diputuskan dengan mempertimbangkan kondisi pasien dan ketersediaan sumber daya. Pendekatan bedah dan obat dengan progesteron yang diberikan sebelum dan sesudah operasi akan meningkatkan kemungkinan luaran pasien dan kehamilan intrauterine yang baik.

**Kata kunci:** kehamilan, heterotopik, laparotomi, progesteron, reseksi kornual, sumber daya rendah.

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## INTRODUCTION

Heterotopic pregnancy (HP) is a rare condition when intrauterine and extrauterine gestations coexist. The incidence of spontaneous heterotopic pregnancy is rare affecting approximately 1 in 30,000 pregnancies. Meanwhile the reported incidence of HP increases from 1:100 to 1:500 with the use of assisted reproductive technology (ART)<sup>1</sup>. The most common ectopic site is the fallopian tube, both in spontaneous and ART heterotopic pregnancies. The second most site is the cornual section. Meanwhile, HP in the cervix, ovary, and abdomen is extremely rare<sup>2</sup>. The diagnosis and management of HP is challenging. Early diagnosis is often complicated due to the co-existence of intrauterine pregnancy (IUP). Delayed diagnosis can result in increased rates of morbidity and mortality both for the mother and IUP. The goal of management of HP is to terminate the ectopic pregnancy (EP) while minimizing the risks towards the IUP<sup>3</sup>. Here we present a rare case of heterotopic pregnancy with threatened abortion of IUP in a natural conception.

## CASE

A 34 year-old primigravida with history of 8-9 weeks amenorrhea came to Obstetrics Emergency Room with chief complaint of vaginal bleeding and lower abdominal pain one day before admission. The complaint is associated with nausea and vomit, general weakness, epigastric pain, and bloated stomach. The patient has history of yellow-green vaginal discharge with dyspareunia for the last 3 months.

Further information suggested, pregnancy occurred naturally, no history of both previous pregnancy and an ectopic pregnancy.

On admission, the patient was hemodynamically unstable. Her blood pressure was 80/60 mmHg with heart rate of 115 beats per minute. Other vital signs were within normal limit. The physical examination revealed tenderness on lower-right abdomen. Speculum examination revealed bluish proximal vaginal until the cervix with vaginal discharge came from external orifice of the uterus. Bimanual examination revealed cervical motion tenderness. On ultrasonography examination, an intrauterine pregnancy measuring 8 weeks and 1 day with fetal heart rate of 132 beats per minute and crown rump length of 1.74 cm (Figure 1). A hyperechoic structure with an internal hypoechoic structure was noted in right adnexa, suggesting

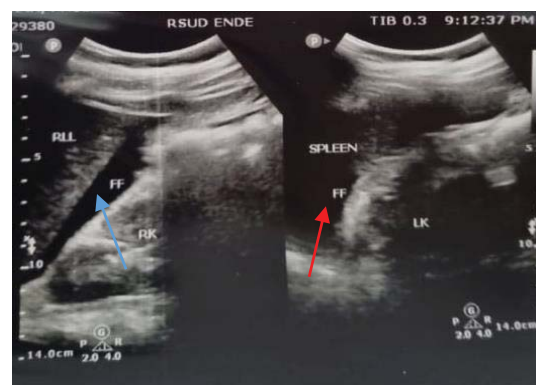
a gestational sac in right uterine horn (Figure 2). There were free fluid in hepatorenal space, splenorenal space, and pouch of Douglas (Figure 3). Laboratory results were notable for positive hCG test, anemia (Hb 10.8 g/dL), and leukocytosis (13.500/ $\mu$ L).



**Figure 1.** Longitudinal transvaginal ultrasound demonstrates an intrauterine gestational sac (red arrow) with a fetal pole (blue arrow). Crown rump length measures 1.74 cm which correlates to the gestational age of 8 weeks and 1 day. Fetal heart rate was 132 beats per minute.



**Figure 2.** Longitudinal transvaginal ultrasound demonstrates a hyperechoic structure with an internal hypoechoic structure noted in right adnexa (red arrow), suggesting a gestational sac in right uterine horn.



**Figure 3.** Transabdominal ultrasound demonstrates free fluid in hepatorenal space (blue arrow), splenorenal space (red arrow), and pouch of Douglas, suggesting occurrence of hemoperitoneum.

The diagnosis of heterotopic pregnancy with ruptured EP was strongly suspected. The patient was managed surgically and medically to remove the EP and preserve the IUP. In order to conserve the threatened IUP, the patient was given one unit of blood transfusion and 200mg micronized progesterone orally 2 times daily before the surgery. Right cornual resection by laparotomy was performed to terminate the EP and the IUP was allowed to continue. The intraoperative findings confirmed ruptured right cornual EP (Figure 4). After the surgery, the progesterone dose was increased to 200 mg 3 times daily. Vaginal swab was conducted to determine the etiology of infection causing vaginal discharge. *Trichomonas vaginalis* was found in the sample. Therefore metronidazole ovula was administered vaginally for 7 days. The patient was scheduled to repeat vaginal swab at 36<sup>th</sup> week of her pregnancy. The patient then was discharged on postoperative day 5 with IUP viability confirmed. At gestation age of 26 weeks, the patient came for check up and ultrasound monitoring showed normal growth of intrauterine pregnancy.



**Figure 4.** Right cornual section by laparotomy was performed and intraoperative findings confirm ruptured right cornual EP.

## DISCUSSION

Heterotopic pregnancy (HP) occurs when intrauterine and ectopic pregnancies exist at the same time. The reported incidence varies widely from 1 in 100 to 1 in 30,000 pregnancies<sup>1</sup>. Heterotopic pregnancy patients who have a spontaneous conception are rarer than those who underwent assisted reproduction. The estimated incidence of HP in spontaneous pregnancies is 1/7,000 to 1/30,000<sup>3</sup>. An intrauterine pregnancy is detected during ultrasonography examination, and an extrauterine pregnancy may be overlooked, causing delay to HP diagnosis<sup>1</sup>.

The notable risk factors for the occurrence of a heterotopic pregnancy include family history, a history of extrauterine pregnancy, previous surgery (including salpingectomy, salpingostomy, or reconstructive tubal surgery), endometriosis, tubal disease, history of pelvic inflammation, high hormone levels, embryo transfer technique<sup>4,5</sup>. Our patient conceived naturally and had one of the risk factors which is history of pelvic inflammation caused by *trichomoniasis*. Tubal pregnancy is the most common location, while interstitial, cornual, and cervical ectopic pregnancies were less frequent<sup>5</sup>. Our case was located on cornual part, which makes our case less common.

The most common symptoms of HP are abdominal pain, vaginal bleeding, adnexal mass, peritoneal irritation and uterine enlargement<sup>6</sup>. Late detection may evolve towards hemoperitoneum following rupture of the EP, and cause hypovolemic shock to the mother<sup>7</sup>. Our case presents with similar signs and symptoms which are lower abdominal pain and vaginal bleeding. The patient was also suspected of rupture of EP because of the unstable hemodynamic and severe abdominal pain.

The first-line adjunct examination is abdominal and transvaginal ultrasound to confirm the diagnosis of both pregnancies. It could also help to evaluate viability of the intrauterine pregnancy and the site of the ectopic pregnancy<sup>7</sup>. In our case, both intrauterine and ectopic pregnancies were visible by ultrasound.

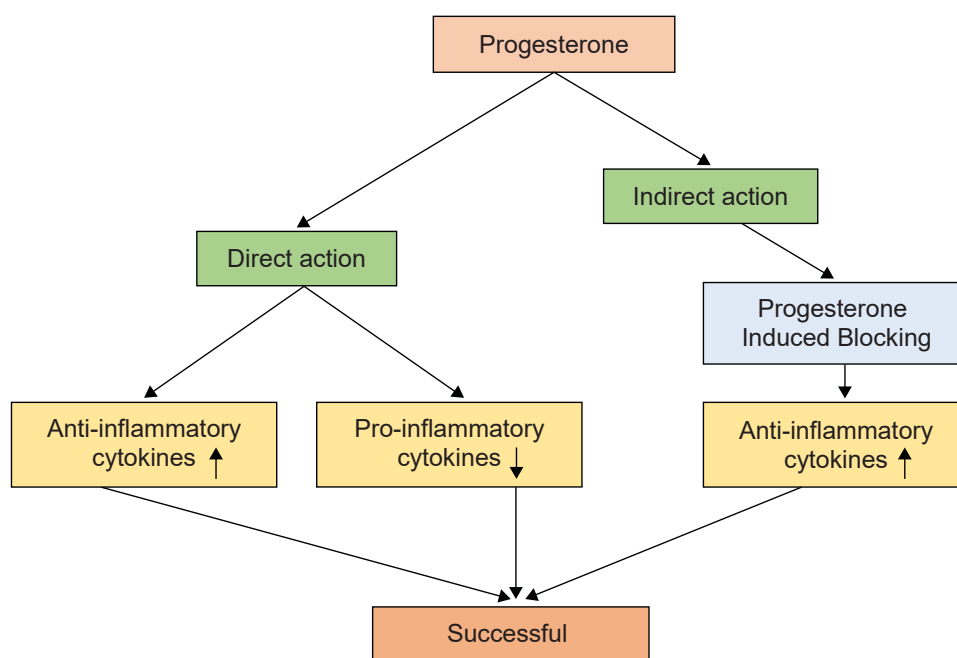
The goal of management of HP is to terminate the ectopic pregnancy while minimizing the risks to IUP. In addition to that, it aims to preserve the patient's fertility and avoid recurrence<sup>7</sup>. Treatment approach for HP should be executed surgically and medically as early as possible. Treatment depends on the patient's condition, the size and site of an EP, previous pregnancies, the viability of intrauterine and extrauterine gestation, and the expertise of the physicians<sup>8</sup>. For patients with unstable hemodynamics or with any signs indicating the rupture of extrauterine pregnancy, emergency surgery is strongly indicated. The benefit of surgical treatment is the ability to completely eliminate the EP, though there might be a higher risk of abortion of the IUP. In their study found that the total abortion rate was 26.56% in all HP patients and the abortion rate in surgery management group was 25.93%<sup>9</sup>. The surgery can be conducted with laparoscopy or laparotomy. Laparoscopy has the advantage of avoiding the risk of uterine manipulation,

compared to laparotomy, which can increase the risk of spontaneous abortion. However, laparotomy is indicated in cases of hemodynamic instability or large hemoperitoneum<sup>7</sup>.

In our case, we combined surgical treatment to remove the EP and medical supplementation to preserve the IUP. Cornual resection by laparotomy method was chosen as the surgical option rather than laparoscopy, considering the hemodynamic instability condition of our patient and the unavailability of laparoscopic resources in our hospital that is located in rural area. The surgery was successful with normal growth of IU embryo. About 60–70% of HP cases result in live childbirth with outcomes similar to that of singleton pregnancies<sup>8</sup>. In order to help preserving the viability of the IUP and decrease the risk of its spontaneous abortion, we administered postsurgical progesterone supplementation to the patient.

Progesterone is often termed as “pregnancy hormone”, as it functions to prepare endometrium for the implantation as well as gestational sac maintenance in the uterus

throughout early pregnancy<sup>10</sup>. It was found that for women who have both bleeding in the first trimester and a history of previous miscarriage, progesterone can have benefit in reducing the risk of miscarrying a fetus<sup>8</sup>. There were some possible mechanisms behind antiabortive effects of progesterone in early pregnancy. It was reported that progesterone functions as an immunomodulator that shifts the maternal cytokine balance from a Th1 or pro-inflammatory bias towards a Th2 or anti-inflammatory bias<sup>11,12</sup>. Thus, in our case it was hoped the administration of progesterone supplementation will reduce inflammation caused by surgical wound and prevent threatened abortion of the IUP (Figure 5). Similar case as ours prescribed vaginal progesterone postsurgical for luteal phase support<sup>8</sup>. They reported good outcome of the IUP. However, we chose to administer progesterone orally as it was found that oral management was demonstrated to be more effective and have a lower risk of miscarriage compared with vaginal administration<sup>10,11</sup>.



**Figure 5.** Possible mechanisms of progesterone in reducing inflammation and preventing abortion<sup>12</sup>

## CONCLUSIONS

Heterotopic pregnancy is a rare condition. Despite a rather challenging condition, early diagnosis and prompt treatment are required to improve outcome both for the patient and the pregnancy. The treatment approach should

be considered based on the patient's condition and availability of resources. Surgical approach and medical approach with progesterone given before and after the surgery would likely improve the outcome of the patient and the intrauterine pregnancy.

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