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Tubaric Pregnancy Combined an Intrauterine Pregnancy with Placental Abruption: A Rare Clinical Case in Emergency and Review of the Literature

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doi: https://doi.org/10.37745/bjmas.2022.04927

Published July 12, 2025

Citation: Zuccon W., Viganò G., Susca M., Padrenostro M., Siliprandi V. (2025) Tubaric Pregnancy Combined an Intrauterine Pregnancy with Placental Abruption: A Rare Clinical Case in Emergency and Review of the Literature, British Journal of Multidisciplinary and Advanced Studies,6(4)15-21

Abstract: A tubaric pregnancy combined with an intrauterine pregnancy with placental abruption is an extremely rare and very high-risk condition that requires urgent and highly specialized medical care. The coexistence of these two conditions makes the diagnosis difficult, because symptoms can often overlap and there is a high risk of bleeding, which is why it requires a high index of suspicion. Risk factors can be multiple but most frequently involve the use of assisted reproductive technologies. Treatment options, from observation to surgery, must be carefully selected based on the clinical situation and management must aim at improving the obstetric outcome for a viable intrauterine pregnancy. Surgical treatment is aimed at removing the tubaric pregnancy, controlling the bleeding and preserving the intrauterine pregnancy. For patients with unstable hemodynamics or with any signs indicating rupture of an ectopic pregnancy, emergency surgery is always strongly recommended. Coexisting placental abruption in an intrauterine pregnancy, if it is partial as in the reported clinical case, may reabsorb spontaneously. The maternal and intrauterine fetal prognosis depends on early diagnosis and fetal survival is good if diagnosis is timely, treatment is appropriate and strict follow-up is performed. At this stage, close monitoring in hospitalization is required and emotional support is essential because the situation is extremely stressful for the patient.

Key words: Tubaric pregnancy, Intrauterin pregnancy, Placental abruption.

INRODUCTION

Clinical Case

A 33-year-old woman presented to our Emergency Department complaining of epigastric pain radiating to the pelvis with nausea and light vaginal bleeding, without vomiting, fever or urinary disorders, the bowel movement was patent with semi-formed stools. She was pregnant with a gestational sac of 8 weeks + 5 days of gestation with known placental abruption for which she was taking progesterone therapy in ovules and folic acid. The past anamnesis was positive for cholecystectomy, tonsillectomy and psoriasis, as well as for

British Journal of Multidisciplinary and Advanced Studies,6(4)15-21, 2025 Health and Medical Sciences Print ISSN: 2517-276X Online ISSN: 2517-2778

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previous pharmacological therapy with synthetic ovulation stimulants. At the gynecological evaluation, the dark red blood loss was scant while the intrauterine gestational sac showed a single viable embryo of 18.9 mm and an area of failed amnio-choral attachment at the lower pole of 28×11 mm, without fluid layers in the Douglas space. Complete blood tests were substantially normal except for an increase in inflammation indices (Wbc 14.81 10^9/L – Neutrophil Granulocytes 11.62 10^9/L / 78.5% - C Reactive Protein 4.64 mg/dl). Due to the persistence of pain, the subsequent transabdominal ultrasound revealed hepatic steatosis and the presence of a complex formation of approximately 6 cm in the right adnexal without fluid collections in the abdomen (**Figure 1**).



Figure 1. Right tubaric pregnancy.

A few hours later, under observation and monitoring, the patient had a clinical worsening with acute abdominal pain, hypotension (98/60 mmHg) and anemia. In fact, the blood tests were normal except for a further increase of the neutrophil leukocytosis (Wbc 24.56 10^{9} /L - Neutrophil Granulocytes 21.99 10^{9} /L / 89.6%) and a reduction in the values of hemoglobin (Hgb from 12.4 gr/dl to 9.4 gr/dl), erythrocytes (Rbc from 4.06 10^{12} /L to 3.08 10^{12} /L) and hematocrit (Hct from 37.2% to 28.4%), while the transabdominal and transvaginal ultrasound, compared to the previous ones, documented the appearance of abundant fluid effusion localized in the perihepatic area, between the loops and in the pelvis with a right annex of still increased volume (**Figure 2**).

British Journal of Multidisciplinary and Advanced Studies,6(4)15-21, 2025 Health and Medical Sciences Print ISSN: 2517-276X Online ISSN: 2517-2778

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Figure 2. Hemoperitoneum due to ruptured tubaric pregnancy.

With a suspected diagnosis of hemoperitoneum, the patient was a candidate for emergency surgery, with blood bags available, exploratory laparoscopy, drainage of hemoperitoneum and right salpingectomy, with preservation of intrauterine fetal viability.

There were no complications in the post - surgical period and the patient was discharged on the third post - operative day in good health.

DISCUSSION

The first case of combined intrauterine and extrauterine pregnancy was reported by Duvernay in 1708 and, according to the most recent statistics, simultaneous pregnancies of this type occur with a theoretically estimated incidence of 1 in 30,000 natural conceptions, but may be more frequent in women who undergo ovulation induction or assisted reproductive techniques¹⁻³.

The natural and spontaneous synchronous development of a second tubaric extrauterine pregnancy during an intrauterine pregnancy with placental abruption is, as in the reported case, an extremely rare and clinically complex situation. In general, it could fall within heterotopic pregnancy, or the simultaneous coexistence of a natural intrauterine pregnancy and an extrauterine one, a condition already rare in itself and in any case more common in cases of assisted fertilization; in fact, although spontaneous cases continue to occur during natural conception, the frequency of cases of heterotopic pregnancy has constantly increased with the use and development of assisted medically reproduction⁴. It can represent a potentially lethal condition that can lead to particular complications such as the rupture of an ectopic pregnancy with hemorrhage and the loss of the intrauterine embryo after treatment⁵.

Risk factors include a history of infertility, sexually transmitted infections, salpingitis, pelvic inflammatory disease or PID, in women with an intrauterine device, smoking, use of hormonal contraception or pelvic surgery, as more frequently in the case of ovarian stimulation for the treatment of infertility or use of assisted reproductive technologies⁶. The fallopian tube is the most frequent site of ectopic implantation in heterotopic pregnancies, but other extrauterine sites may also be involved simultaneously, such as the cervix and, exceptionally, the ovary, the peritoneum or the abdominal cavity. In multiple pregnancies, two fertilized ovules during the same cycle implant and develop simultaneously, one in the uterine cavity, the other in an

British Journal of Multidisciplinary and Advanced Studies,6(4)15-21, 2025 Health and Medical Sciences Print ISSN: 2517-276X Online ISSN: 2517-2778

https://bjmas.org/index.php/bjmas/index

extrauterine location. Regarding the origin of the two fertilized ovules, belonging to the same cycle, it is possible that they come either from a single follicle originally provided with two egg cells or from two follicles that matured at the same time, located in the same ovary or in two different ovaries^{7,8}.

The simultaneity of a multiple pregnancy often makes the diagnosis difficult because symptoms, such as abdominal pain or bleeding may overlap, and the 'falsely reassuring' presence of an intrauterine fetus may be the cause of a delay in early diagnosis and timely treatment, conditions necessary at interrupting the ectopic pregnancy and minimizing damage to the mother and intrauterine fetus; in this regard, involving a high risk of haemorrhage, both for tubaric pregnancy and for placental abruption, clinical management must include close monitoring in hospital with frequent ultrasound scans, serial blood counts and assessment of intrauterine fetal viability^{9,10}.

Symptoms may present with abdominal pain, localized or diffuse, associated with vaginal bleeding or spotting, as well as palpation of an adnexal swelling. Depending on the stage of the disease, where 50% of cases present with rupture and hemorrhage with hemoperitoneum and subsequent hypovolemic shock, the patient may be hypotensive or hemodynamically unstable. There are no specific physical or laboratory tests for heterotopic pregnancy, but this diagnosis should be considered in any pregnant patient with abdominal pain, hypotension, and an ultrasound examination demonstrating free fluid, as well as a history of assisted medically procreation^{11,12}.

As reported in the literature, transvaginal ultrasound is better in early diagnosis than transabdominal ultrasound, detecting almost 70% of cases between the fifth and eighth week of gestation, but in almost half of the cases this problem is diagnosed during emergency surgical exploration, while in about 60-70% of cases it ends with a live birth^{13,14}.

Management of waiting depends on many variables such as gestational age and patient conditions, size and location of the ectopic pregnancy, and other risk factors. In relation to the high failure rate, waiting may be selected in asymptomatic patients and in whom the unruptured ectopic embryo has a limited cranio-caudal length without cardiac activity. Instead, ultrasound-guided embryo aspiration should be performed only when the ectopic gestational sac is clearly visualized¹⁵.

For patients with unstable hemodynamics or any signs indicating rupture of an ectopic pregnancy, emergency surgery is always strongly recommended¹⁶. Surgical management is aimed at removing the tubaric pregnancy, controlling the hemorrhage and preserving the intrauterine pregnancy, where careful manipulation of the uterus is essential during procedures for fetal survival. Laparoscopic surgery is a safe and effective treatment without increasing the risk of miscarriage or neonatal malformations; in fact, surgery performed in good time, even laparoscopically, allows a good prognosis. After the surgical phase, rest, hydration and monitoring of vital parameters are essential and any blood loss, signs of infection or recurrence of abdominal pain should be monitored to try to predict post-operative complications^{17,18}.

Placental abruption in intrauterine pregnancy, if partial, as in the reported clinical case, may be spontaneously reabsorbed; in this phase, clinical management includes complete rest, the possible use of progesterone, measurement of β -hcg levels and frequent transvaginal ultrasound monitoring to assess embryonic viability. The use of progesterone for women who have vaginal bleeding in the first trimester, a history of previous spontaneous abortion or a threatened abortion, may have beneficial effects in reducing the risk of spontaneous abortion of a fetus¹⁹.

British Journal of Multidisciplinary and Advanced Studies, 6(4) 15-21, 2025

Health and Medical Sciences

Print ISSN: 2517-276X

Online ISSN: 2517-2778

https://bjmas.org/index.php/bjmas/index

In the reported clinical case, the extrauterine embryo implanted in the fallopian tube and this condition caused the subsequent rupture of the tube and internal bleeding with a risk to the mother's life. The concomitant placental abruption of the intrauterine pregnancy, which however appeared to be progressing normally, could complicate the clinical and surgical management since, where the placenta separates prematurely from the uterine wall compromising the supply of oxygen and nutrients to the fetus, it could represent an additional condition of hemorrhage and risk not only for the mother but also for the fetus. The heterotopic pregnancy, diagnosed and treated promptly, did not have unfavorable outcomes for either the intrauterine pregnancy or the woman, and in this phase the emotional support to the patient was essential because the situation is extremely stressful.

CONCLUSION

Heterotopic pregnancy, or combined pregnancy, is an obstetric phenomenon that involves significant maternal morbidity and mortality due to the risk of rupture of the ectopic pregnancy. The simultaneous development of a uterine pregnancy and an ectopic pregnancy is still a rare occurrence and worthy of particular interest.

The coexistence of these two conditions, tubaric pregnancy and placental abruption, also involve a difficult diagnosis because the symptoms can be misleading, involves a double risk of haemorrhage for both the tubaric pregnancy and the placental abruption, therefore requiring intensive monitoring in hospital. Transvaginal ultrasound can provide the diagnosis which can then be confirmed by laparoscopic exploration in order to allow treatment of the ectopic pregnancy.

All pregnant women presenting with acute abdominal pain or bleeding and in case of hemorrhagic shock, especially after assisted medically procreation, should be suspected of heterotopic pregnancy and undergo surgical exploration, although the clinical differential diagnosis is often not easy.

Therapeutic options include different methods, from observation to surgery, and must be carefully selected according to the clinical situation, where the diagnosis requires a high index of suspicion and management aimed at improving the obstetric outcome for a viable intrauterine pregnancy. In fact, maternal and fetal intrauterine prognosis depends on early diagnosis and there are good chances of having a viable uterine fetus if the diagnosis is timely, if the treatment is appropriate and if a rigorous follow-up is performed, although further studies are needed to optimize treatment protocols.

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British Journal of Multidisciplinary and Advanced Studies, 6(4) 15-21, 2025

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Print ISSN: 2517-276X

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Authors' Contribution

WZ and VS: Planned work, conducted the literature search and produced the report.GV and MS and MP: Helped in contributed to produce the report.WZ and VS: Final revision before delivery.All Authors have reviewed the final report and approve its publication.

Conflict of interest

This work has no financial support by the creator or other sources. None of the authors have any conflicts of interest to disclose.

Ethical considerations

Support for this work has been entirely provided by institutional and / or departmental sources. Partecipants' data will be provided after approval by the Related Author and the Health Management of the Major Hospital, Crema.

Acknowledgement

Eleonora Valori (Gynecology and Obstetrics Unit), Alberto Longo and Pierluigi La Piana (Surgery Unit).

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