Management of abdominal pregnancy with placenta left in situ

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Introduction: Abdominal pregnancy is a rare form of ectopic pregnancy with a challenging diagnosis and management. We reported two cases of abdominal pregnancy with the placenta left in situ, which had different outcomes.

Case Presentation: Case one was a woman (P1A1) referred due to active bleeding after an emergency laparotomy for advanced abdominal pregnancy (36–37 weeks gestational age [GA]). The newborn (3300 g) was delivered alive, but the placenta was left in situ due to hemodynamic instability. Re-exploration surgery revealed a placenta in the abdominal cavity, covering the caecum and ileocaecal junction. The placenta was left due to high suspicion of infiltration into the retroperitoneum and hemodynamic instability. Serial β-hCG levels declined progressively but increased again at day 48. A methotrexate regimen (20 mg/day) for five days was given, and the level was 6.03 mIU/ml on day 94. However, the abdominal wound was still seeping even after 12 months post-operatively due to suspicion of fistula formation. The placenta was fully absorbed after 14 months post-operative. Case two was a woman (G2P1) referred due to ectopic pregnancy and fetal demise at 14-15 weeks GA. Emergency laparotomy revealed abdominal pregnancy. The placenta was also left intra-abdominal due to active bleeding in the implantation area. The serial β-hCG declined progressively, with no neovascularization, and the placenta was fully absorbed four months and six months after the operation, respectively.

Conclusion: In managing abdominal pregnancy with placenta left in situ, periodic quantitative β-hCG, ultrasound examination of the placental size, and monitoring of surgical wounds are indispensable. Chemotherapy must be initiated if there is a plateau or an increase of serial β-hCG level or neovascularization of placental Doppler.

Keywords: abdominal pregnancy, β-human chorionic gonadotropin, left placenta in situ, methotrexate, emergency.

CASE PRESENTATION

Case One

Mrs. B, a 34-year-old woman (P1A1), was referred to Dr. Soetomo General Hospital's Emergency Room for further management of active bleeding after emergency laparotomy due to advanced abdominal pregnancy with placenta left in situ. She had a history of infertility, miscarriage, and curettage. There was no history of tubal surgery, ectopic pregnancy, or in vitro fertilization. At 34-35 weeks GA, she was diagnosed with placenta previa and planned operation at 37-38 weeks GA. No suspicion of abdominal pregnancy was made.

Two weeks later (at 36-37 weeks GA), the patient came to a local hospital due to severe abdominal pain with clinical signs of shock. Emergency laparotomy revealed 2,000 ccs of bleeding in the abdominal cavity. The fetus was delivered by foot extraction, female, 3,300 g, with an APGAR score of 3-5-7. The right and left
CASE REPORT

Mrs. M, a 36-year-old woman (G2A1), was referred to Dr. Soetomo General Hospital’s Emergency Room for further management of abdominal pregnancy with fetal demise. She had no history of infertility, previous miscarriage, or ectopic pregnancy. During the first trimester of pregnancy, she received antenatal care from a local midwife but had no history of ultrasound. At 14-15 weeks GA, the patient complained about abdominal pain 1 day before admission to a local hospital. Ultrasound by a local obstetrician revealed extrauterine pregnancy. The uterus was anteflexy and had a visible endometrial lining. There was no fetal heartbeat, so the patient was referred to Dr. Soetomo General Hospital’s Emergency Room for further management of abdominal pregnancy with fetal demise. She had no history of infertility, previous miscarriage, or ectopic pregnancy. During the first trimester of pregnancy, she received antenatal care from a local midwife but had no history of ultrasound. At 14-15 weeks GA, the patient complained about abdominal pain 1 day before admission to a local hospital. Ultrasound by a local obstetrician revealed extrauterine pregnancy. The uterus was anteflexy and had a visible endometrial lining. There was no fetal heartbeat, so the patient was referred to our hospital.

Upon arrival, an ultrasound was done, revealing free fluid in douglas’, splenorenal and hepatorenal pouches. Explorative surgery was done in collaboration with the digestive surgery department on the day after referral. The evaluation revealed the placenta in the abdominal cavity with an anterior-facing fetal side. The size was approximately 15 x 20 cm wide, covering the caecum and ileocaecal junction. Although it was difficult to evaluate, the surgical team highly suspected that the placenta was attached to the retroperitoneum, so we left it in situ. Then, the placenta was tied with suede thread, and the surgeon gave some abdominal tampons due to placental implantation bleeding. Subsequently, the abdomen was closed and given an uphill drain before the patient was referred.

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After discharge, the patient was followed up regularly in the gynecology outpatient clinic for surgical wound examination, serial β-hCG, and ultrasound to evaluate the placental size and vascularity index. Serial β-hCG on days 3, 7, 11, 15 and 19 post-operative revealed progressive decline (19,115; 11,067; 5,476; 3,074; 3,156.6 mIU/ml, respectively) and reached the hundreds level but then increased again at day 48 due to suspicious of malignancy. Surgical wounds were still seeping on day 82 postoperative. The patient was then consulted at the gynecology oncology outpatient clinics and advised to receive a methotrexate chemotherapy injection of 20 mg/day for five consecutive days. Two weeks after the methotrexate chemotherapy (on day 94 postoperative), the β-hCG levels decreased to 6.03 mIU/ml. Besides the β-hCG levels, the ultrasound evaluation of placental size also decreased along with time, and no increase in vascularization was found. The abdominal wound was still seeping at 12 months post-operative due to suspicion of fistula formation. There was no sign of infections based on the patient’s clinical findings and CBC (Cell Blood Count) result. Ultrasonography follow-up of placental size and vascularization (using Doppler velocimetry) revealed that the placental was fully absorbed after 14 months post-operative. Two years after surgery, the abdominal wound defect was fully closed without any complaints, and now the patient is trying to get pregnant.

Case Two

Mrs. M, a 36-year-old woman (G2A1), was referred to Dr. Soetomo General Hospital’s Emergency Room for further management of abdominal pregnancy with fetal demise. She had no history of infertility, previous miscarriage, or ectopic pregnancy. During the first trimester of pregnancy, she received antenatal care from a local midwife but had no history of ultrasound. At 14-15 weeks GA, the patient complained about abdominal pain 1 day before admission to a local hospital. Ultrasound by a local obstetrician revealed extrauterine pregnancy. The uterus was anteflexy and had a visible endometrial lining. There was no fetal heartbeat, so the patient was referred to our hospital.

On admission to our hospital, she was pale, slightly hypotensive, and had tachycardia. Internal examination revealed...
a cervical opening of 1 cm, tenderness of the portion, an enlarged uterus, and a prominent Douglas’ cavity. Ultrasound examination revealed an extraterine fetus (biometry 14/15 weeks GA), no heartbeat, and free fluid in the Morrison’s and Douglas’ pouch (Figure 2). The patient was also anemic (Hb 7.3).

Emergency laparotomy found abdominal pregnancy connected to the right adnexa with implantation of the placenta extending from the outside of the fundus–posterior uterine - cervix and small intestine. The right and left tubes and ovaries were challenging to evaluate. The gestational sac was delivered, revealing a blood clot of 400 ccs and a fetus weighing 100 g (Figure 3). There was active bleeding in the area of placental implantation that was difficult to evaluate, so we chose to leave the placenta intact. We placed 6 abdominal tampons and an uphill drain and then closed the surgical field. The patient received antibiotic therapy and was re-operated to remove the abdominal tampon after 2x24 hours from the first operation.

Patients were educated to perform quantitative β-hCG examination and ultrasound periodically to determine the size of the placenta. The patient was discharged after 7 days. There was a progressive decline in serial quantitative β-hCG examination on days 0, 29, 49, and 72 postoperative (4,407; 275.9; 32; 1.65 mIU/ml, respectively). The surgical wound was still seeping and closed completely after the 3 months post-operative. The size of the placenta decreased at four months postoperatively, and there was no neovascularization around the implanted placenta. The ultrasonography result during follow-up showed that the placenta was fully absorbed 6 months postoperative. The abdominal wound was also in good condition without any complaints. After 2 years post-operative, the patient was not yet pregnant.

**DISCUSSION**

We reported two cases of abdominal pregnancy that we managed in our center. Our center is a tertiary-level hospital with 622 live births annually in 2021. In both cases, the patient was referred from type B and C hospitals, abdominal pregnancy diagnoses were made intraoperatively during surgery, and both were managed with placenta left in situ. However, both cases had different fetal outcomes and maternal recovery trajectories.

Firstly, diagnosing abdominal pregnancy is very challenging and requires a high index of suspicion. There was no pathognomonic sign of abdominal pregnancy. The patient could be asymptomatic or have symptoms like nonspecific abdominal pain. The pain can be progressive as the pregnancy progresses, accompanied by other symptoms such as painful fetal movements or gastrointestinal disturbances. Furthermore, intraabdominal bleeding complications may present as acute abdomen with or without clinical signs of shock. In our cases, both patients came to the emergency room with acute abdomen and hemodynamic instability.

Diagnosis of abdominal pregnancy can be made through imaging or visualization of extraterine pregnancy in the presence of an intact uterus. In both of our cases, diagnoses were made intraoperatively. However, in the first case, the diagnosis of AAP was made intraoperatively even though the patient had routine antenatal care with an obstetrician since the first trimester of pregnancy. Nkusu et al., on their case report and review of 163 AAP cases reported since 1946, found that most AAP cases (55%) were diagnosed intraoperatively. These were understandable due to the complexity of its diagnosis through ultrasound. Some proposed major ultrasound features that may help a physician to notice intraabdominal pregnancy include evidence of a fetus in a gestational sac outside the uterus, difficulty or failure to visualize the uterine wall between the baby and bladder, the close proximity between the fetus and the abdominal wall, and demonstration of placenta out of the uterus. The recognition of these features should prompt physicians to refer patients for further imaging, such as magnetic resonance imaging, to confirm the diagnosis and map the extent of placental involvement and its vascular supply to make a sound treatment plan.

Next, managing abdominal pregnancy needs a multidisciplinary care team, various treatment options, and the availability of blood bank services. It all depends on many factors (e.g., hemodynamic stability, fetal status (i.e., alive versus demised), and the gestational age (e.g., ≤20 or >20 weeks at which the abdominal pregnancy was diagnosed). Patients with hemodynamic instability or embryonic or fetal demise require surgical removal of the pregnancy, performed by laparotomy or laparoscopic surgery. Selective arterial embolization may be performed in some patients before surgical management to reduce blood loss.

In hemodynamically stable patients with live gestations, there was no evidence-based management strategy, patients may choose expectant management or termination, and the decision is generally determined by gestational age. We perform surgical management for hemodynamically stable patients with gestations ≤20 weeks or >20 weeks who chose termination. The evidence of adjuvant therapies such as injection of potassium chloride, selective arterial embolization, or methotrexate is of limited efficacy; however, adjuvant therapies may be used in selected patients (e.g., patients in whom the abdominal pregnancy has colonized vessel-rich areas [such as the liver or spleen], patients with pregnancies >20 weeks of gestation) to help prevent intraoperative hemorrhage. The optimal management of the placenta is unclear. Various management methods have been described in the literature, such as removing the placenta in surgery, leaving the placenta in situ, selective embolization, and pre-or postoperative methotrexate. In cases where the placenta is challenging to be removed, it may be left in situ. However, this approach may predispose patients to adverse events, such as hemorrhage, sepsis, disseminated coagulation, bowel obstruction, and fistula formation. In such cases, postoperative adjuvant therapies (e.g., selective arterial embolization, methotrexate, mifepristone) may be used to induce degradation of the placenta and decrease the risk of intraabdominal hemorrhage. However, postoperative methotrexate can lead to rapid placental lobular necrosis, resulting in intraabdominal abscesses, bleeding, and even death.
was an advanced abdominal pregnancy with a live fetus, while the second was an early abdominal pregnancy with fetal demise. In both cases, we managed by laparotomy surgery and left the placenta in situ because of a massive hemorrhage. We didn’t give these patients routine methotrexate injections after surgery because of its adverse events.1,17

In managing abdominal pregnancy with the left placenta in situ, some factors must be examined periodically to prevent some adverse events. A few days after surgery, we observe some adverse events that may develop, such as secondary hemorrhage, sepsis, disseminated coagulation, or bowel obstruction. After discharge from the hospital, we periodically examine the β-hCG quantitative levels, ultrasonography of placental size, Doppler ultrasonography of placental blood supply (vascular index), and monitoring of surgical wounds. In our cases, the first case showed an increase in β-hCG quantitative levels after 48 days of surgery and the leakage of surgical wounds after 82 days, even after 12 months postoperative.18 We managed this patient with a methotrexate injection of 20 mg/day for 5 days in a row, and two weeks later, the β-hCG levels dropped significantly. Our second case showed no rise in β-hCG quantitative levels approximately 4 months after surgery. The placenta decreased in volume and fully absorbed after 6 months.

Some literature stated different conclusions about how long it takes for the placenta to be completely absorbed. Cetinkaya et al. reported an 83% reduction of the placental volume by the end of 1-year observation and a significant decrease in β-hCG level after three months.19 Another report by Rohilla et al. reported that spontaneous placental resorption would happen at a maximum of 15 months, and embolization of the feeding vessel may be considered to accelerate placental autolysis.1 In two cases of abnormally adherent placenta reported by Torrenga et al., the serum β-hCG level normalized within 10 weeks, whereas the placental regression happened slowly up to nine months after delivery.19 The two cases we present are very different in terms of gestational age, and of course, the placenta size is very different. This may affect the duration of placental resorption and the normal return of the β-hCG quantitative levels. Even though the β-hCG can return to normal, persisting symptoms can justify surgical removal of placental remnants for patients’ better quality of life.15,20

The two cases that have been reported involve patients who were referred due to emergency issues. In response, urgent actions were taken, which can often present challenges in determining the most optimum approach to treatment. The outcomes of the cases also vary. There is a necessity for a meticulously performed case report on abdominal pregnancy, which would enable the identification of differences in the outcomes observed in these types of situations.

CONCLUSIONS

The diagnosis and management of abdominal pregnancy confront significant challenges. In the management of abdominal pregnancy with the placenta retained in situ, it is essential to do regular assessments of quantitative β-hCG levels, perform ultrasound examinations to monitor placental size, and closely observe surgical wounds. If there is a plateau or even an increase in the level of β-hCG quantitatively, chemotherapy should be initiated to prevent a detrimental outcome to the patient. There is a positive correlation between the duration needed for complete absorption of the placenta and the gestational age; the higher the gestational age, the longer the time of placental absorption.

CONFLICT OF INTERESTS

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AUTHOR CONTRIBUTION

SI: data collection, data analysis, manuscript draft. EE: conceived research idea, supervised manuscript draft and review. HE, MY: co-supervised manuscript review. All authors read and approved the manuscript.

ETHICAL CONSIDERATION

A subject voluntarily consented to the study’s publication, understanding that the patient’s identity would remain private. ICMJE (International Committee of Medical Journal Editors) ethics approval has been obtained.

REFERENCES


