

A successful management of an advanced secondary abdominal pregnancy with a live fetus: A case report*

BY KATERINE MAE FERNANDO, MD; CARMENCITA B. TONGCO, MD, FPOGS AND SHERYL ANN B. DELA CRUZ, MD, FPOGS
Department of Obstetrics and Gynecology, East Avenue Medical Center

ABSTRACT

Advanced abdominal pregnancy is associated with catastrophic outcomes for both mother and fetus. Because it is rare, it is often misdiagnosed and the surgery, often unplanned, may end up with uncontrollable hemorrhage and injury to abdominal structures during placental removal. A case of a 21-year-old G1P0, 34 weeks gestation, who presented as a bleeding placenta previa but diagnosed intraoperatively as abdominal pregnancy with a live baby with congenital anomalies, with complete removal of the placenta and with good maternal outcome is presented. This report highlights the pitfalls in diagnosis and stresses the importance of team management, adherence to good surgical principles, and timely operative decisions to ensure a successful outcome when preoperative evaluation is not possible.

Keywords: advanced abdominal pregnancy, ectopic pregnancy, live baby, placental removal

INTRODUCTION

An ectopic pregnancy that implants within the peritoneal cavity but excluding the fallopian tubes, ovaries, and broad ligament is called an abdominal pregnancy. While approximately 90% of ectopic pregnancies are tubal, only 1.4% are abdominal, with an estimated incidence of 1 in 10,000 to 25,000 live births¹. Based on the Philippine Obstetrical and Gynecological Society annual report from 2008-2018, abdominal pregnancy accounted for approximately 0.43% of all ectopic pregnancies². Records from our institution from 2013-2018 show that there was only one report of an abdominal pregnancy (8 weeks gestation) located on the vesico-uterine pouch.

An advanced abdominal pregnancy with a viable fetus is therefore, a rare phenomenon. Because it is uncommon, and the signs and symptoms are non-specific, it is most often misdiagnosed and discovered only during laparotomy. Thus, the condition is associated with high maternal and perinatal mortality and morbidity³. Furthermore, standard management guidelines do not exist and the approach is largely individualized. This case report documents the first advanced abdominal pregnancy that reached 34 weeks gestation in our institution over the past five years,

albeit misdiagnosed as placenta previa, yet was delivered successfully with minimal maternal morbidity and with a live baby. This report aims to discuss possible strategies to improve diagnosis and more importantly, to stress the importance of team management, adherence to good surgical principles, and timely operative decisions which ensured a successful outcome even when preoperative evaluation was not possible.

CASE REPORT

A 21-year-old primigravid was admitted at our institution at 34 weeks and 1 day age of gestation for placenta previa. The medical history and pregnancy course were unremarkable except for an episode of UTI at 6 months AOG. Three days prior to admission, with vaginal spotting, hypogastric pain, and an ultrasound diagnosis of previa totalis and oligohydramnios, she was referred to our institution. At our emergency room, the patient was in pain but with normal vital signs and not pale. Abdomen was globular but non-tender. Fundic height was 27 cm, in breech presentation, with good fetal heart tones. Occasional strong uterine contractions were palpated. On speculum, the cervix was closed, with minimal bleeding.

The plan was to do cesarean section after steroid administration, and to procure blood products. However, on the twelfth hour, the patient bled and this prompted immediate surgery. An intraoperative diagnosis of abdominal pregnancy was made. A 20 cm placenta was on the left, with the fetus lodged within the right paracolic

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gutter, in breech, with no amniotic sac (Figure 1). There was no hemoperitoneum.

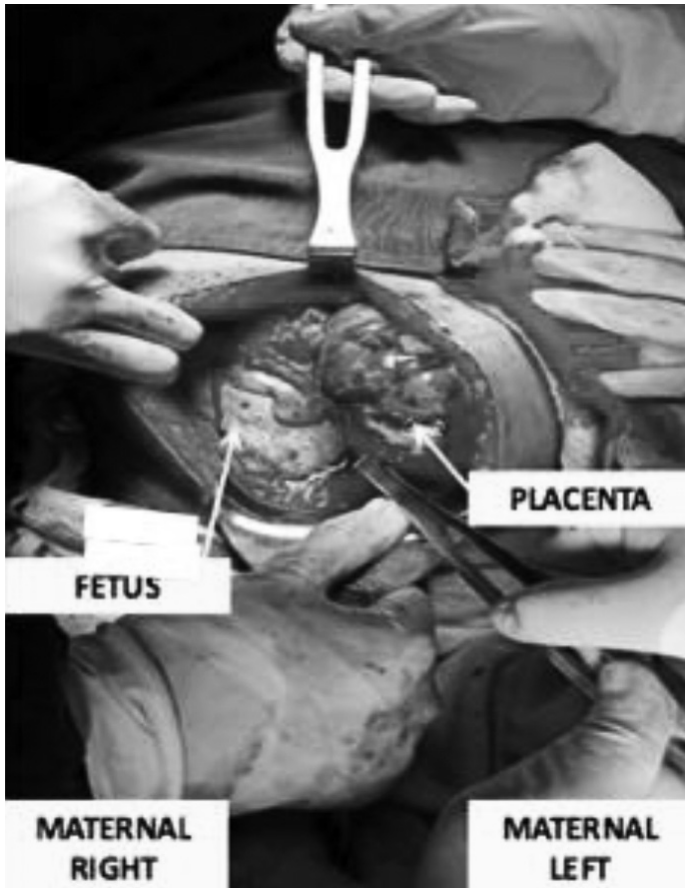


Figure 1. Intra-operative findings showing fetus and placenta

Anticipating a difficult surgery, we sought the assistance of a more senior surgeon. A second large bore intravenous line was inserted and 4 units of packed red cells were secured before proceeding with the delivery. When ample blood products became available, the operating team proceeded with delivery of the baby by easily scooping the head, which was more accessible than the breech. On further inspection, the placenta was seen implanted superior to the left adnexa and adherent to it and to the posterior uterine wall (Figure 2). The left IP ligament was markedly engorged, with large vessels going to the placenta. The rest of the pelvic organs were normal. A left salpingoophorectomy and lysis of placental attachments to remove the placenta were done. The patient tolerated the two and a half hour procedure well, with blood loss of 700 ml. She was discharged on the third hospital day and counseled regarding the importance of early documentation of her next pregnancy.

The baby was asphyxiated and subsequently expired after 15 minutes of life from respiratory failure due to pulmonary hypoplasia. She had signs of Potter syndrome (Figure 3).

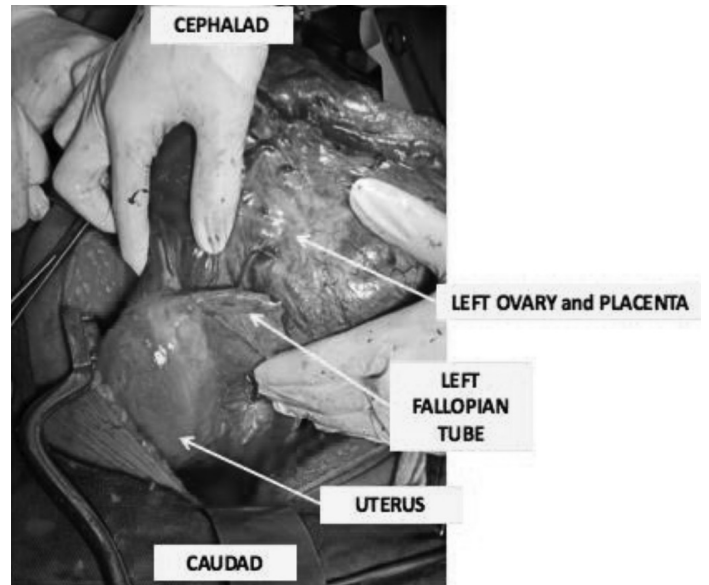


Figure 2. Uterus was slightly enlarged with no areas of bleeding or rupture. The left fallopian tube and ovary were encased within the placenta



Figure 3. The baby with Potter facies: low set ears, prominent epicanthic folds and downward slant of eyes expired, clubbed feet, small compressed thorax and chest wall

The final diagnosis was:

G1P1(0100) Abdominal Pregnancy, advanced, secondary, delivered, preterm, breech, live birth
S/P exploratory laparotomy with extraction of the fetus, and removal of the placenta after left salpingoophorectomy and adhesiolysis
Early neonatal death secondary to respiratory failure from pulmonary hypoplasia of Potter syndrome
Blood transfusion (2 units packed red cells)

DISCUSSION

An ectopic pregnancy that implants within the peritoneal cavity is called an abdominal pregnancy.

Abdominal pregnancy is classified as 1) primary or secondary, or 2) early or advanced. An abdominal pregnancy is secondary when the fetus continues to grow in the abdominal cavity after being extruded from the fallopian tube or the uterus. Our patient did not fulfill the Studdifort's criteria¹ for primary abdominal pregnancy which includes: 1. presence of normal bilateral tubes and ovaries with no evidence of recent or past pregnancy; 2. no evidence of a uteroperitoneal fistula; and 3. presence of pregnancy related exclusively to the peritoneal surface, early enough to eliminate the possibility of secondary implantation after primary tubal nidation. Classifying an abdominal pregnancy as early (<20 weeks gestation) or advanced (>20 weeks gestation) has more clinical relevance because it may influence decisions as to timing of intervention in relation to fetal viability and as to how the placenta should be managed⁴.

Our patient has a secondary and an advanced abdominal pregnancy. The longer the pregnancy, the bigger the placenta, with its propensity for bleeding and greater morbidity. Her abdominal pregnancy probably started as an early tubal rupture or abortion, with subsequent implantation of the placenta close to the site of rupture, with the fetus having more room for growth in the right paracolic gutter. The pregnancy reached 34 weeks because of the generous blood supply from the left ovarian artery to the placenta.

Why did we miss the diagnosis? Many of the symptoms are non-specific and variable depending on the location of the pregnancy⁴. Our patient experienced pain and bleeding only on admission. Rupture of her amniotic sac should have caused severe pain due to peritoneal irritation, yet she was largely asymptomatic. Instead of placenta previa, which is painless bleeding, a more plausible diagnosis would have been abruptio placenta or uterine rupture because our patient came in pain⁵.

We failed to suspect abdominal pregnancy because of the absence of risk factors in our patient like Pelvic Inflammatory Disease (PID), previous uterine surgeries, dilatation and curettage, previous tubal pregnancy and artificial insemination⁴.

We did not appreciate the usual findings in an abdominal pregnancy because her fetus was deep in the right paracolic gutter and was not easily palpable. But its extremely lateral location should have made us suspect an unusual pregnancy. We also misinterpreted the term-sized placenta as the uterus with contractions.

Obviously, we were influenced by the ultrasound report of placenta previa. The sonologist's role is crucial in establishing the correct diagnosis. A routine obstetric ultrasound should involve identification of the cervix as it continues into the myometrium superiorly, an amniotic sac adjacent to the cervix, and the presence of myometrium

between the bladder and the pregnancy. When these are not demonstrated, abdominal pregnancy is highly likely. Other common sonologic features of abdominal pregnancy should be noted (Table 1). In spite of these clues, diagnostic errors with ultrasound as reported in literature are high, ranging from 50% to 90% especially for advanced abdominal pregnancies because of confounders like bowel loops, fibroids, a retroflexed position, a uterine horn, and pregnancy in a bicornuate uterus⁴.

Table 1. Sonographic Features in Abdominal Pregnancy

Sonographic Features in Abdominal Pregnancy ⁷	
	No of Patients (%)
Uterus and fetus separate	18 (90)
Ectopic placenta	15 (75)
Oligohydramnios	9 (45)
Fetal parts in close proximity to maternal wall	5 (25)
Abnormal lie	5 (25)
Poor visualization of placenta	5 (25)
Maternal bowel gas blocking visualization	5 (25)
Failure to visualize myometrium between fetus and maternal bladder	3 (15)
Failure to visualize myometrium between placenta and maternal bladder	3 (15)
Placenta previa appearance	3 (15)
Maternal peritoneal fluid	2 (10)

Magnetic Resonance Imaging (MRI) is emerging as a complementary tool to ultrasound in detecting abdominal pregnancy. Also, MRI with contrast will map out the vascular and placental organ invasion which are important for preoperative planning on how the placenta should be managed. When the placenta invades large vessels like the aorta and vena cava, the liver, and spleen, and omentum, it might be more prudent to leave the placenta behind to avoid excessive blood loss⁴.

The two most important issues to discuss in the management of our case with advanced abdominal pregnancy are: the timing of delivery for fetuses nearing viability and the management of the placenta. Because advanced abdominal pregnancies are not common, management guidelines are not yet established.

Experience from case reports favor immediate termination in abdominal pregnancy regardless of fetal age, because of the risk of fatal intra-abdominal hemorrhage. It is amazing how this patient's abdominal pregnancy was carried to near term without maternal complications, in spite of the lack of appropriate obstetrical care. There are case reports, although limited,

where the decision to delay surgery for a short period resulted in viable fetuses (Table 2) but under very strict conditions like: a hemodynamically stable patient, strict

Table 2. Case Reports of Advanced Abdominal Pregnancy with viable fetus

Author	Summary of the journals with viable fetus	Outcome
Silva et al. ⁸	A 36 years old G2P2 diagnosed with abdominal pregnancy at 18 2/7 weeks of gestation. A supraumbilical laparotomy was performed at 34 weeks age of gestation for fetal extraction.	A female newborn weighing 2187grams, with an Agar score of 5/8. Baby was discharged after 2 weeks of medical treatment of necrotizing enterocolitis.
Gidiri et al. ⁹	A 37 years old G2P0 35 4/7 weeks AOG baby in oblique lie, buttocks under the liver, head in epigastrium and a placenta attached to fundus of uterus externally.	Normal male 2-kg baby was delivered
Huang K. et al. ³	Intrauterine pregnancy combined with an abdominal pregnancy. The abdominal pregnancy placenta was attached to the lower end of the uterine wall. Diagnosed at 26 weeks AOG. Pregnancy was terminated by cesarean section at 33 5/7 weeks gestation.	<u>Pregnancy uterine:</u> Apgar score of 9 at 1 minute and 10 at 5 minutes, and weighed 1990g without deformities. <u>Abdominal pregnancy:</u> Fetus sat with a single hip inside the sac. When newborn was delivered an an Apgar score of 0 at 1 minute and 0 at 5 minutes, and weighed 1600 g. The placenta was posterior to the uterine wall, covering the upper anterior sigmoid colon and the rectum

in-hospital monitoring, 24 hour access to anesthesia, surgery, and blood products, available ultrasound to detect early signs of abdominal gestational sac rupture, congenital anomalies, monitoring of fetal well-being, and MRI to evaluate the placenta. From an ethical standpoint, this option of conservative management should adhere to bioethical principles of patient autonomy, beneficence and non-maleficence, with the end directed towards the greatest benefit for the patient and her baby.

After the diagnosis of abdominal pregnancy is made and if the patient is stable, prompt preoperative planning should be undertaken, An extensive and thorough patient and family counseling is ideal. Consultations with other specialties like anesthesia, neonatology, surgery, urology, hematology, interventional radiology and vascular surgery are vital and they should form part of the management team. An MRI with contrast will help plan the appropriate management for the placenta. Adequate blood products are secured in anticipation of bleeding.

What is the best way to manage the placenta? There are two possible options: 1) removal, either partial or complete, or 2) conservative, that is, leaving it in-situ to avoid hemorrhage during removal. Conservative management is combined with embolization of the feeding vessel after surgery to hasten placental involution. The advantages and disadvantages of both approaches are seen on Table 3. The decision on placental management has to be individualized depending on the location and extent of the placenta, the patient's condition at surgery, the surgeon's expertise, availability of blood products, and access to ancillary treatment like embolization. These factors were considered in managing the placenta of our patient. She is young and we needed to preserve her fertility. The placenta was limited in location and minimally adherent. She was hemodynamically stable and blood was ready. We did not have access to embolization. Therefore, we decided that placental removal is the appropriate choice.

We performed a systematic inspection of the abdomino-pelvic cavity: The 20 cm diameter placenta, with its prominent vasculature, was noted to be superior to the pelvic organs, more to the left, with the left fallopian tube and ovary adherent to and encased by placenta (Figure 2). It also extended to the area of the posterior uterine wall and was adherent to it. The uterus was slightly enlarged and had a smooth serosa. The right fallopian tube and ovary were grossly normal. The cul-de-sac peritoneum, urinary bladder, small bowels, and omentum were uninvolved. The next step was to identify the blood supply of the placenta and to ligate it prior to proceeding with lysis of adherent areas. The blood supply of the pelvis was traced, starting from the sacral promontory at the aortic bifurcation and down to the

common iliac and its branches on the pelvic sidewall. If ligation of the hypogastric arteries was necessary, we were already able to map out its location. We noted the left infundibulopelvic ligament to be swollen and engorged, and this was where the placenta was receiving its blood supply. A cardinal rule to minimize blood loss during surgery is to ligate the source of the blood supply to the pathology to be removed. Thus, the left ovarian artery was ligated, followed by the left uteroovarian ligament to cut off the blood flow from the area of anastomosis of the ovarian and uterine arteries lose to it. These steps minimized blood loss during dissection of the placenta from its adherent sites. Electrosurgery was employed. The estimated blood loss was 700ml, necessitating transfusion of only two units of packed red cells.

We delivered a live baby girl weighing 2050gm with Potter syndrome: low set ears, prominent epicanthic folds and downward slant of eyes, clubbed feet, small compressed thorax and chest wall (Figure 3). The baby

expired after fifteen minutes of life due to respiratory failure due to pulmonary hypoplasia. The rupture of the amniotic sac and loss of amniotic fluid lead to compressive symptoms and pulmonary hypoplasia⁶. Does abdominal pregnancy predispose to congenital fetal malformation? Reports are conflicting. Fetal malformations can be as high as 40% due to compression and absence of amniotic fluid⁶. The most common defect is congenital foot malformation followed by joint contractures, facial asymmetry, mild spasticity and intrauterine growth restriction.

There is paucity of information regarding the future reproductive potential of patients who have had abdominal pregnancy. It can be surmised that fertility potential may be proportional to the damage to the fallopian tubes and ovaries³. Our patient's left fallopian tube and ovary needed to be removed because these were markedly adherent to the placenta and dissection might lead to bleeding. Nevertheless, her right fallopian tube and ovary are normal and so her chances for future

Table 3. Management of the Placenta in Advanced Abdominal Pregnancy

TECHNIQUE	Comment	Advantages	Disadvantages
Complete evacuation	Morbidity is lower if blood supply is stopped either by ligation or embolization, prior to evacuation.	Treatment is complete, no need to monitor for involution	May be bloody, prone to injuries to neighboring organs adherent to the placenta.
Partial evacuation	Avoids excessive hemorrhage or injury if difficult removal is expected	Prevents further blood loss	Need to monitor for involution; risk of infection, necrosis, fistula, bowel obstruction
Non-evacuation	An alternative If evacuation is unsafe.	Minimizes bleeding and injury to neighboring organs where placenta adheres to; need to ligate the cord as close to the placenta as possible.	Greater postoperative morbidity compared with removal; risk of infection, fistula, ileus, peritonitis, bowel obstruction, abdominal pain, intermittent fever, abscess formation, toxemias of pregnancy and late hemorrhage
Embolization	An adjunct to radical or conservative management, either preoperatively to decrease bleeding during evacuation, or postoperatively to facilitate placenta resorption.	Minimally invasive	Complications of ileus, sepsis and intestinal perforation, arterial bleeding if faulty technique.
Medical management: methotrexate (systemic and local) ⁶ , local instillation of potassium chloride, hyperosmolar glucose, prostaglandins, danazol, etoposide and mifepristone ^{3,4} .	Facilitates placental absorption	Non-invasive	Limited studies; may need repeated use, together with BHCG monitoring; risk of infection, bleeding ³ .

pregnancy may be good, granting that these will not be affected by adhesion formation. She has to be counseled regarding the importance of early documentation of her next pregnancy to avoid the delays which endangered her life in this current pregnancy.

SUMMARY

Advanced abdominal pregnancy is associated with catastrophic outcomes for both mother and fetus because of late diagnosis and operative complications like bleeding and injury to neighboring structures, confounded by lack of adequate preoperative preparation. Because advanced abdominal pregnancy is rare and the manifestations are

variable, it is difficult to diagnose and in most cases, the diagnosis is made during laparotomy. The role of the sonologist is very crucial in the diagnosis. If there is a high index of suspicion for abdominal pregnancy, in spite of normal ultrasound findings, a repeat ultrasound should be requested. Surgery, the mainstay of management, should be tailored to placental extent, the risk for hemorrhage and infection, the surgeon's expertise, and available facilities (embolization/ blood bank). If time permits, a thorough preoperative workup is pivotal in planning the management for the placenta. The importance of team management, adherence to good surgical principles, and timely operative decisions can never be overemphasized. ■

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