



RESEARCH ARTICLE

EXTRA UTERINE ABDOMINAL PREGNANCY WITH A LIVE FETUS: A CASE REPORT

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ABSTRACT

Extrauterine pregnancy is an extremely rare condition in which the fetus develops outside the uterine cavity and instead in the maternal abdominal cavity. This article aims to report a case assisted by the advanced gestation team with late diagnosis in which exploratory laparotomy was performed to remove a live conceptus and resection of placental tissue from the uterine cavity and the vessels where it was nourished. This is a rare case due to a good maternal-fetal prognosis.

Key words:

Extrauterine Pregnancy; Ectopic;
Laparotomy.

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INTRODUCTION

Ectopic pregnancy (EP) is defined as implantation and development of a pregnancy outside the uterine cavity, being called abdominal ectopic pregnancy (AEP) when it occurs inside the abdominal cavity^{1,2}. EP is detected in high incidence rates in the uterine tubes, corresponding to around 96 to 99% of cases, while AEP is found in only 0.5% of pregnancies, configuring the rarity of the case³. The prevalence of EP has been increasing over the years and represents 2% of pregnancies in the United States, being the fourth leading cause of maternal death in the United Kingdom^{1,2}. It is responsible for 9% of deaths in the first gestational trimester in developed countries, although its mortality rate has decreased over the last decade in these countries. Regarding the prevalence of AEP, it corresponds to one in every 402 gestations in developed countries and one in every 10,000 gestations in developing countries⁴. In Brazil, specifically, there are no regional or even national prevalence measures for

this event⁵. In most of AEP cases, fetal decease is verified due to placental insufficiency, as a result of poor implantation of the trophoblast^{6,7}. Deformities of the limbs and face of the fetus, as well as pulmonary hypoplasia, resulting from compression by the abdominal structures, are also common^{2,7}. The maternal morbidity and mortality rates are high^{8,9,10}, and cataclysmic complications are frequent, such as hemorrhage of the placental bed⁶, often caused by difficulty in placental extraction, as a consequence of its implantation in noble viscera, like the liver, and it is not uncommon for the medical team to decide to keep the placenta in situ, in order to avoid profuse bleeding and injury to abdominal structures^{6,11}. Abdominal pregnancy with a viable fetus is also an even greater rarity, with only a few sporadic cases being described in the literature. Neonatal survival is about 20% in the largest published series^{7,8,10}. In Brazil, the greatest experience with abdominal pregnancy was described by Costa et al. in Recife, PE^{12,13,14}, but on few occasions a live fetus was found with neonatal survival conditions^{12,14}. In 1992, Alencar et al.¹⁵ described a case of advanced abdominal pregnancy with a live

newborn at the Assis Chateaubriand Maternity School, in Fortaleza, CE. Mendonça et al.¹⁶, in João Pessoa, PB, also reported, in 1993, a case of rupture of an advanced abdominal pregnancy with a live fetus, which progressed with severe hemorrhage. Due to its extreme uniqueness, it is therefore recommended to disclose all cases that come to be diagnosed.

In the present study, the authors reported their experience with a case of late abdominal pregnancy conducted at the São Vicente de Paulo Hospital and Maternity (SVPHM) in the municipality of Barbalha, in the countryside of Ceará, obtaining a live conceptus and satisfactory evolution, with maternal and neonatal survival.

CASE REPORT

G.M.B.S, 34 years old, G4 P1-0-2-1 (1 vaginal delivery, 2 spontaneous abortions in the first trimester), from Juazeiro do Norte, CE, admitted to SVPHM on April 16th, 2024, with a history of abdominal pain since the beginning of the current gestation, which worsened with fetal movement, intensifying in the last three months of pregnancy. Her gestational age estimated according to the last menstrual period, 08/05/2023, was of 36 weeks and 3 days, and according to an ultrasound performed on 01/08/2024, was of 34 weeks and 6 days.

This exam showed topical pregnancy at 20 weeks and 5 days. She had a prenatal card with 6 consultations without serious complications, only complaints of dysuria, pain in the right iliac fossa and nausea; and two laboratory routines with no significant changes, only anemia, with hemoglobin of 10.4 (on 01/19/2024). She had an ultrasound performed on 03/08/2024 showing a single fetus, in longitudinal lie, left position and breech presentation, with fetal heart rate of 123 bpm, fetal weight of 951g (1st percentile for gestational age), an homogeneous placenta, of high insertion, classified as Grannum's grade 0, with amniotic fluid of reduced volume with the deepest vertical pocket of 1.1 cm, and presence of a uterine nodule located in the anterior wall, measuring 9.9 cm in length and 7.9 cm in width.

She also underwent Pelvic Magnetic Resonance Imaging, without contrast, on 04/12/2024 to evaluate the placental mesenchymal mass, evidencing pregnancy in an extrauterine location, occupying the maternal abdominopelvic cavity, characterized by a single fetus in a longitudinal situation, breech presentation and dorsum on the left, with no myometrium identified covering the gestational structures (placenta and chorioamniotic membranes),

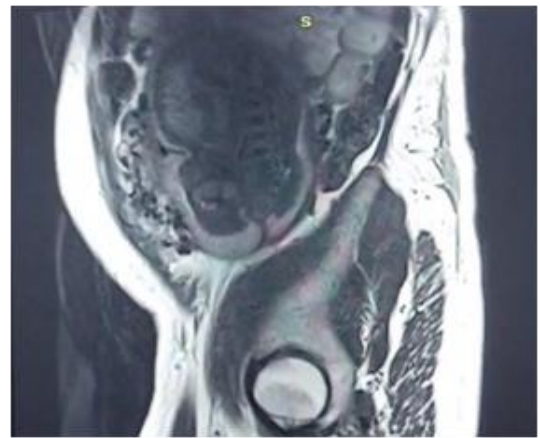
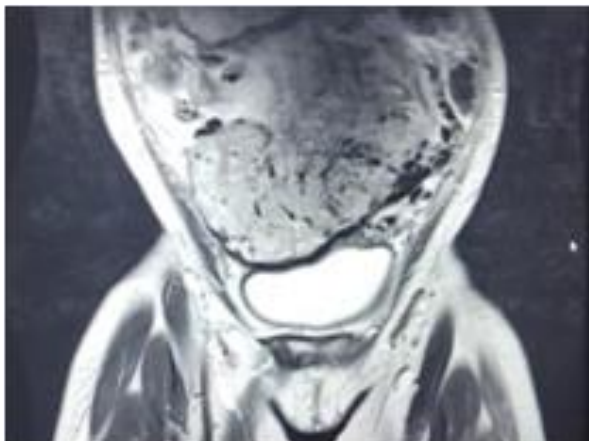


Figure 1. MRI showing placenta and fetus in the abdominal cavity

heterogeneous placenta, in anterior location, with prominent intraplacental vessels and hypointense bands on T2-weighted sequences, average placental thickness of 7.5 cm, showing anatomical relations with the adjacent maternal structures, immediately posterior to the rectus abdominis muscles in the mesogastrium/hypogastrium region (its upper end located about 3.0 cm above the level of the umbilical scar), exhibiting extensive contact with those muscle bellies and even insinuating itself through the umbilical scar, not ruling out some degree of adhesion/invasion of the anterior abdominal wall; it presented extensive contact with the bladder domus, with multiple prominent vascular structures at the interface between the placenta and the bladder, some degree of adherence to the outer layer of the bladder was not ruled out; there was no evidence of transmural bladder involvement; it displayed ample contact with the external iliac vessels on the right; Multiple prominent vessels were seen surrounding the placenta, especially on its lateral and inferior surfaces, as well as in the vesicouterine recess. On admission physical examination, the patient was in regular general condition, pale (++)/4+, malnourished, borderline hydrated, afebrile, anicteric, acyanotic, without edema. Blood pressure was 100/60 mmHg, heart rate was 98 bpm, and cardiopulmonary auscultation showed no abnormalities. The abdomen was painful on superficial and deep palpation, establishing the superficiality of the fetal parts, with an abdominal height of 27 cm. The fetus was in a longitudinal situation, and it was not possible to determine the presentation. The fetal heart rate (FHR) was 138 bpm, auscultated in the left hypochondrium. The cervical exam revealed a firm, posterior and impervious cervix. The diagnostic hypotheses suggested at the hospitalization were advanced ectopic pregnancy with a live fetus, anemia and malnutrition. Blood count, blood classification, coagulation tests and obstetric ultrasound were requested. On the same day, the obstetric ultrasound with transvaginal complement was performed, with the following report: single fetus, longitudinal situation, breech presentation, female dorsum to the left, fetal weight of 2484g (54th percentile when correlating with the first ultrasound), Amniotic Fluid Index of 1.0 cm, FHR of 150 bpm, anterior placenta difficult to characterize in terms of placental degree, heterogeneous, thickness of 79 mm. Doppler velocimetry exhibited large vessels permeating the mass, with normal ratio in flowmetry of umbilical artery (Pulsatility Index 0.62; Resistive Index 0.48) and middle cerebral artery (Pulsatility Index 1.20; Resistive Index 0.71). The transvaginal ultrasonography showcased a retroverted uterus measuring 9.9 cm in length, 7.7 cm in width and 8.0 cm in depth, with 311

cm³ in volume. The ultrasonographic study reported abdominal pregnancy with live fetus at 34 weeks and 5 days of gestational age and impaired fetal vitality due to oligohydramnios.



Figure 2. Ultrasonography images showing placental mass and vascularization on Doppler study

Considering the gestational age of 34 weeks and 6 days by the earliest ultrasound, the decrease in amniotic fluid, and the maternal-fetal risk intrinsic to this type of pregnancy, exploratory laparotomy was indicated for extraction of the conceptus and placenta, being preceded by detailed preoperative care. The patient was put on a liquid diet without residues on April 20th, and on a fasting period on April 21st, 24 hours before the procedure, under fluid infusion of Ringer's Lactate and hypertonic glucose IV solution. She also received blood transfusion with 2 (two) units of packed red blood cells on 04/21/2024 since she had hemoglobin values of 8.1 g% and hematocrit of 24.4%; Correction of hydroelectrolyte disturbances was performed with intravenous potassium restitution (value of 2.9 mg/dL) and magnesium restoration due to hypomagnesemia (1.8 mg/dL). Post-transfusion hemoglobin was 10.4 g%, and hematocrit was 31.4%. Another 4 (four) units of packed red blood cells, 3 (three) units of plasma and 2 (two) units of platelets were set aside for the patient. Coagulation, liver and kidney function tests, and electrocardiogram with surgical risk assessment were also checked (with evidence of low risk for the proposed surgical procedure). Constant surveillance of fetal vitality and maternal hemodynamic conditions was maintained throughout the preoperative period. Exploratory laparotomy was performed on 04/22/2024, and the patient underwent usual monitoring with pulse oximetry, 5-lead cardioscopy, non-invasive blood pressure, and active heating with a thermal blanket under the patient. On spontaneous ventilation with supplemental oxygen through a 3 L/min nasal catheter, sedated in Ramsay 2 with 2 mg of midazolam by peripheral venoclysis for puncture of the Right Subclavian Vein with a Double-Lumen Central Venous Catheter under local anesthesia with 5 mL of 2% Isobaric Lidocaine. With the patient seated, epidural block was performed with an 18G Tuohy needle at the T9-T10 level by the Pending Drop Technique (or Gutierrez's) technique, in an

aseptic manner, a solution of 18 mL of ropivacaine 2mg/mL without epinephrine and 2 mg of morphine was injected. The patient was placed in supine position and volume expansion was performed with heated crystalloids. Nursing team performed the passage of an Indwelling Urinary Probe with an efflux of a small amount of diuresis, somewhat concentrated. The procedure began with a median xipho-umbilical incision, observing the child in the amniotic sac with a minimal amount of fluid. The live female fetus was quickly removed. After fetal extraction, pre-oxygenation was performed and conversion to general anesthesia with anesthetic induction and rapid sequence orotracheal intubation under direct laryngoscopy and upper airway aspiration.



Figure 3. Fetus perceived in abdominal cavity immediately after the initial incision

After clamping of the umbilical cord, arranged longitudinally with the pelvic pole in the lower abdomen region, and after orotracheal intubation of the patient, the incision was enlarged to the pubic region for a complete inventory of the abdominal cavity. During the incision, part of the placenta and material of the amniotic sac were noted adhered to the parietal peritoneum, and, from the umbilical scar, it was carefully proceeded with the opening, releasing the adhesions. On evaluation of internal conditions, an extremely voluminous placenta was observed that infiltrated multiple organs, including part of the greater omentum, mesocolon of the descending colon, sigmoid colon and transition with the rectum, and part of the cecum. (Figure 4).

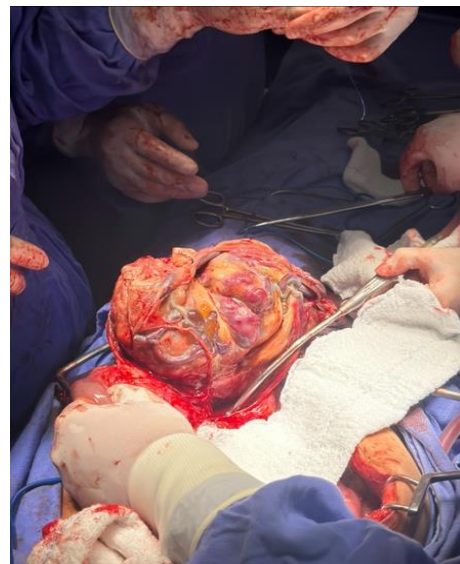


Figure 4. Voluminous placental mass

There was infiltration of the mesentery of the distal jejunum and ileum. In addition, adhesions were also found in the left ureter and left iliac vessels (iliac artery and external iliac vein). Firm adherence and infiltrative aspect were observed in the bladder peritoneum, ovary and left fallopian tube, possibly encompassed by the placental mass, in addition to infiltration in the right tube. Careful dissection of the placenta, a large and extremely vascularized tumor mass, with calibrous and friable vessels that caused bleeding even with the use of hemostatic forceps, was performed. During the dissection of the mesocolon and mesentery, there were arterioles emerging from the placenta towards the vessels of these structures, suggesting nutrition of the conceptus by the placenta. Careful dissection, ligation of the arterioles, and preservation of the intestinal loops were performed. After ureterolysis of the left ureter, the external iliac vessels were released, and the firmly adhered bladder peritoneum was resected. However, for complete resection of the placenta, it was necessary to perform left salpingoophorectomy and right salpingectomy. The next steps of the procedure were rigorous hemostatic review, counting of compresses and materials, and synthesis by planes. Intraoperatively, 3000 mL of crystalloids and 2 units of red blood cell concentrates were infused into the patient. After surgery, the patient was extubated without intercurrents and transferred to the ICU for intensive postoperative care, with central venous access in the right subclavian vein (punctured in the operating room) and with 100 mL diuresis in the indwelling urinary catheter collector, axillary temperature of 35.0°C, heart rate of 70 bpm, respiratory frequency of 16 bpm, pulse oximetry saturation of 99% (without supplemental oxygen), Capillary Blood Glucose of 122 mg/dL, Systolic Blood Pressure (SBP) of 86 mmHg and Diastolic Blood Pressure (DBP) of 59 mmHg (Mean Arterial Pressure of 68 mmHg). She was admitted to the ICU still under residual anesthesia, with Ramsay of 3 (compatible with Glasgow Coma Scale value of 14). She did not present any discomfort when she arrived at the ICU. It was initiated parenteral prokinetic medication (bromopride 10 mg) three times a day. Diuresis in the first 12 hours was only 300 mL; with a positive hydric balance of $\pm 3,000$ mL; with hemodynamics maintained at the expense of vasoactive amine (noradrenaline solution 10 ml/h = ± 0.26 mcg/kg/min). Volume replacement on demand was increased (the patient was already at 2,000 mL in 24 hours); another 1,000 mL of Saline solution 0.9% and then 20 mg of parenteral furosemide (IV) were instilled, with a good response. On the 1st Postoperative Day (POD), a liquid diet was also started with good acceptance and Domperidone 10 mg orally three times a day. At the beginning of the 2nd POD, the patient already had a diuresis of 1,800 mL in 24 hours and a hemodynamic maintained at the expense of a very low dose of norepinephrine solution (5 ml/h = ± 0.12 mcg/kg/min.); the vasoactive amine being weaned and withdrawn during the day. At the beginning of the 3rd POD, she was already without hemodynamic support, accepting the oral diet well and with good diuresis, being discharged from the ICU to the Obstetrics Ward. A preterm female newborn was born by cesarean section by laparotomy, indicated by an intra-abdominal fetus, at 35 weeks and 5 days of gestational age, weighing 2250 grams, 32.5 cm of head circumference, 29 cm of thoracic circumference and 42 cm of height. She was born with good vitality, in breech presentation, without the need for resuscitation maneuvers. APGAR status 9/9, at the 1st and 5th minute of life. On physical examination, the newborn presented cranial asymmetry (plagiocephaly), asymmetry in the face and hip with postural deviation, without other

morphological alterations. The patient was referred to the neonatal ICU due to mild dyspnea estimated at 2 points by the Silverman-Andersen Bulletin, requiring the use of continuous positive airway pressure (CPAP) in the first 12 hours of life. Chest X-ray was consistent with transient tachypnea of the newborn. During hospitalization, antibiotics were empirically initiated, given the risk of neonatal infection, history of oligohydramnios, and the clinical picture after birth. She underwent 3 days of gentamicin, 1 day of ampicillin, and 2 days of procaine penicillin, and antibiotic therapy was suspended after a complete blood count with a score of zero on the Rodwell score and normal CRP in 2 collections, at 12 and 72 hours of life, in addition to blood culture without bacterial growth. Routine transfontanelar and urinary ultrasonography were performed without alterations. At 1 day of life, she was evaluated by orthopedics, which showed mild dysmetria in the right lower limb, which could not rule out developmental dysplasia of the hip, and it was suggested that an ultrasound of the joint be performed on an outpatient basis. After the first 8 days of hospitalization in the intensive care environment, the newborn was transferred to the nursery of the service, where she remained stable and exclusively breastfed. The patient was discharged from the hospital with her mother at 10 days of age, weighing 2310 grams, with corrected gestational age of 37 weeks, and a prescription for a multivitamin supplement for daily use, in addition to guidelines for outpatient follow-up with orthopedics.

DISCUSSION

In most cases of Abdominal Ectopic Pregnancy, fetal death is almost always present^{17,7,13}, and maternal life may be threatened, during pregnancy and resolution, due to the risk of hemorrhage resulting either from premature and spontaneous placental abruption¹⁷ or from obstetric maneuvers for its extraction during surgery. Maternal mortality described in the literature ranges from 5 to 20%^{8,7,18}. In the study by Atrash et al.⁹, the risk of maternal death in the presence of abdominal pregnancy was 7.7 times higher than in tubal pregnancy and 90 times higher than in topical uterine pregnancy. In the case described, there was a favorable evolution of abdominal pregnancy based on detailed pre, trans and postoperative care, as well as a team composed of an obstetrician and an oncological surgeon, with vascular and urological surgeons on call, intensivists and neonatologists with skill and technical preparation to perform the surgical intervention performed. However, it is noteworthy that this type of result is the exception, and not the rule, in the natural history of abdominal pregnancy, in which it generally results in high rates of morbidity and mortality. In addition to hemorrhage, which is the main cause of maternal morbidity and mortality^{6,8}, other complications have been described, such as infection, intestinal obstruction, amniotic embolism, amniointestinal fistula, and hypofibrinogenesis^{8,6,19,20}.

We suggest that the good results in this case are initially because the placenta, when inserted almost completely into the omentum, sparing the noble structures. In addition to this, the surgery was performed only after balancing the clinical, hemodynamic and hydroelectrolytic conditions of the patient, by performing blood transfusion and keeping several units of packed red blood cells set aside, reducing intraabdominal pressure with diet adjustment 48 hours prior to the procedure. These precautions and Preoperative preparations undoubtedly contributed to the surgery being performed in satisfactory

conditions and success in the outcome of the case. Reports in the literature of survival of the fetus are very rare^{12,13,14,15,16}, since fetal death is extremely common^{8,6,18,21}, resulting from both placental insufficiency and abruption. It is described in case series studies fetuses with growth restriction and usually presenting, at birth, characteristics typical of postdatism, such as parchment skin, long bones and nails, and an aging appearance¹³.

Deformities of the lower limbs and fetal head can also be observed, due to permanent contact with maternal solid structures, in the absence of uterine wall protection and amniotic fluid^{6,10,18}, as oligohydramnios represents a frequent finding. In cases where live births are obtained, neonatal mortality is very high, either due to prematurity, pulmonary hypoplasia or hypoxia¹⁸. Another unusual fact of the case presented here is its late diagnosis, even though the patient had six prenatal consultations in another service and had an ultrasound exam performed in the twentieth week of gestation. Although Martin Jr and McCaul report that only 50% of the cases have a prenatal diagnosis, it should be considered that early diagnosis continues to be the key point to avoid the deleterious consequences of abdominal pregnancy²⁰. Ultrasound usually, according to a case series study, closes the diagnosis, especially when performed in the early gestational, because in advanced pregnancies the diagnosis can be difficult. The main echographic findings characteristic of abdominal pregnancy are the presence of the fetus and placenta in an extrauterine location, fetal parts very close to the abdominal wall, and absence of visualization of the myometrium between the fetus and/or placenta and the mother's bladder¹⁸. Oligohydramnios and anomalous presentations are also frequent, and difficulty in visualizing the placenta is usually found^{22,23}. These findings were reported in the ultrasonography exam performed at our service. Due to all these peculiarities of the case described and assisted by the team of authors, we believe that the publication of the case is relevant to serve as a reference and research for the medical profession on how to proceed in the face of similar cases, since it deviates at several points from the usual evolution of advanced abdominal pregnancies, constituting an extremely rare case regarding satisfactory maternal and fetal evolution.

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