A 27-Year-Old Brazilian Woman with a History of Left Salpingectomy and Late Diagnosis of an Extra-Uterine Intra-Abdominal Pregnancy and Live Birth at 26 Weeks’ Gestation

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Financial support: None declared
Conflict of interest: None declared

Patient: Female, 27-year-old
Final Diagnosis: Abdominal pregnancy
Symptoms: None
Medication: —
Clinical Procedure: —
Specialty: Obstetrics and Gynecology
Objective: Rare disease

Background: Abdominal pregnancy is a special type of ectopic pregnancy, characterized by implantation of the embryo in the peritoneal cavity, with tubal, ovarian, and intraligamentary pregnancies excluded, accounting for approximately 1% of all cases. It was first reported in 1708 after an autopsy and since then numerous cases have been reported, with a current incidence of 1: 10 000 to 1: 30 000 pregnancies.

Case Report: We report the case of a 27-year-old woman, resident of the city of Caxias do Sul, Brazil, with an extra-uterine pregnancy by ultrasound diagnosis at 25 weeks and 1 day of gestational age and a live fetus.

Conclusions: Abdominal gestation is a rare type of ectopic pregnancy and is characterized as a life-threatening situation. Its biggest challenge is to make an early diagnosis, since most cases go unnoticed at the ultrasound performed in the first trimester, and when symptomatic, they do not present themselves in a specific way. When necessary, MRI has been shown to greatly elucidate such cases. Moreover, the therapeutic decision also presents some disparities in the literature. Although it is known that open surgery is best option, there are still many doubts regarding whether to perform placental extraction since its removal process can cause abundant bleeding, putting the patient at risk during the surgical procedure, in the same way that its maintenance and the use of drug treatment can also aggravate the patient’s clinical picture.

Keywords: Pregnancy, Abdominal • Pregnancy, Ectopic • Pregnancy, High-Risk

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/934401

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

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Background

Abdominal pregnancy is a special type of ectopic pregnancy, characterized by implantation of the embryo in the peritoneal cavity, with tubal, ovarian, and intraligamentary pregnancies excluded, accounting for approximately 1% of all cases [1]. It was first reported in 1708 after an autopsy and since then numerous cases have been reported, with a current incidence of 1: 10 000 to 1: 30 000 pregnancies [2]. Therefore, we present this case report of late-diagnosed abdominal pregnancy with a live fetus.

Case Report

A 27-year-old woman underwent the first ultrasound of the pregnancy in an external service to the General Hospital and was referred to the same due to suspicion of extra-uterine pregnancy. Ultrasound imaging showed a fetus, apparently located in the abdominal cavity to the right of the uterus, placenta inserted behind the uterus, absent amniotic fluid, fetus with heartbeat, estimated weight of 670 g, and gestational age 25 weeks and 1 day.

The patient had a history of pregnancy in the left uterine horn, with resection and raffia of the same in 2014, with the tube and ovary preserved. She had no previous comorbidities, except a diagnosis of gestational diabetes with lifestyle change treatment; other prenatal exams within the normal range.

Magnetic resonance imaging of the total abdomen was performed on the day of admission. No topical pregnancy was identified. There was placental formation in the topography of the left adnexal region, surrounded by the ipsilateral fallopian tube. The fetus was inside the abdominal cavity, predominantly located on the right flank and iliac fossa, with the cephalic portion between the bladder and the rectum, and the lower limbs interlocking on the right flank. We found marked pyelo-calicical dilatation with transition in caliber of the ureters at the level of the described alterations (Figures 1-3).

Aiming at lung maturation, 2 ampoules, each with 1 mL containing 3 mg betamethasone acetate and 3.945 mg betamethasone disodium phosphate were injected intramuscularly and a new obstetric ultrasound was performed for a better fetal evaluation, showing an irregular cranial contour. There was a cystic image with thin septa adjacent to the fetal neck measuring 3.5×2.7 cm. The heart had 4 cavities and was apparently balanced. The thorax was apparently reduced in size, and it was not possible to evaluate the integrity of the diaphragmatic dome. The feet were in plantar flexion and the bladder had increased in size.

After a multidisciplinary discussion with the oncologic surgery team (which was invited to participate in the case due to its
complexity) in which maternal and fetal risks and benefits were extensively evaluated, and also after explanation and discussion with the patient, aiming at a better evaluation of placental implantation in large vessels, it was decided to perform an angiotomography with abdominal contrast to ensure a better surgical planning to assure the patient’s safety (Figure 4).

The examination demonstrated increased arterial impregnation of the intrauterine vessels; signs of volumetric increase of the tubal region to the left, with increased vascularization by contrast; placenta located in the hypogastric/iliac fossa to the right with extension to the mesogastric region/left flank, without cleavage plane with posterior body wall of the uterus and left uterine attachment; calibrous vessels from the uterine and left ovarian arteries apparently supplying the placenta were visualized, as well as signs of venous drainage from the placenta through the left gonadal vein, which presented with increased caliber; and there were no signs of involvement of the intestinal loops by the placenta. After performing an angiotomography, the team talked to the patient about interrupting the pregnancy due to the increased risk to the mother’s life as the pregnancy progressed. Vascular surgery, urology, oncology surgery, and obstetrics teams met.

Surgery was performed in 3 stages. Before starting the surgery, spinal anesthesia was performed and a conversion to general anesthesia was planned after delivery. In the first step, the urological surgery team implanted a double-J catheter bilaterally to facilitate identification of the ureters in case more extensive surgery involving hysterectomy was needed during the transoperative period. In the second step, the vascular surgery team performed catheterization of the right femoral artery with balloon placement in the left hypogastric artery to reduce major bleeding.

In a third and last step, exploratory laparotomy was performed. A fetus was visualized (Figure 5), without amniotic sac, between intestinal loops in the abdominal cavity, removed in pelvic presentation with immediate clamping of the umbilical cord (Figure 6), with the placenta implanted in the left uterine artery and left gonadal artery and posterior uterine wall with invasion of the sigmoid rectum (Figure 7). Due to massive bleeding after fetal removal and the patient’s evolution to hemodynamic instability, uterine preservation and conservative management with placenta maintenance and clinical treatment for absorption were not possible and a monoblock extended total hysterectomy with sigmoid (Figures 8, 9) and colostomy was performed. In the transoperative period, the patient presented hypovolemic shock, requiring replacement...

**Figure 3.** MRI coronal section requested for better elucidation of abdominal ectopic pregnancy, in which the following structures are identified and indicated by the letters in white: P – placenta, F – fetus.

**Figure 4.** Sagittal section of contrast-enhanced CT angiography performed to check for the possibility of vessel invasion by the placenta. In the image we have the following structures identified by the abbreviations in red: P – placenta, AICE – left common iliac artery, AIIE – internal iliac artery, AUE – left uterine artery.
After being moved to the ICU for postoperative recovery, she was discharged to the ward in less than 24 h. The female neonate was immediately attended to by the neonatal ICU team. She presented an Apgar score of 2 in the first minute and 6 in the fifth minute, weighing 960 g, and with a physical examination compatible with 26 weeks and 3 days of gestational age. She required of blood products, with good recovery. After being moved to the ICU for postoperative recovery, she was discharged to the ward in less than 24 h. The female neonate was immediately attended to by the neonatal ICU team. She presented an Apgar score of 2 in the first minute and 6 in the fifth minute, weighing 960 g, and with a physical examination compatible with 26 weeks and 3 days of gestational age. She required of blood products, with good recovery. After being moved to the ICU for postoperative recovery, she was discharged to the ward in less than 24 h. The female neonate was immediately attended to by the neonatal ICU team. She presented an Apgar score of 2 in the first minute and 6 in the fifth minute, weighing 960 g, and with a physical examination compatible with 26 weeks and 3 days of gestational age. She required of blood products, with good recovery.
It is a life-threatening situation due to high maternal and fetal morbidity and mortality, with a maternal mortality rate around 0.5% to 18% and a perinatal mortality rate between 40% and 95% [4]. In addition, it is estimated that approximately 21% of newborns from an abdominal pregnancy have some malformation, probably due to compression of the fetus in the absence of amniotic fluid. Typical deformities include limb defects, facial and cranial asymmetry, joint abnormalities, and central nervous system malformations [5]. However, its early detection is difficult, with only 45% of diagnoses being made prenatally [6], with most cases diagnosed after complications or only intraoperatively. When symptomatic, the clinical signs are not very specific, but some authors believe that some signs can alert to the possibility of the diagnosis: abdominal pain with disordered bowel movements, abdominal pain during active fetal movements, spread of the abdomen due to an irregular presentation, palpation of fetal parts below the maternal abdominal wall, and failure to trigger labor. Unfortunately, these signs only appear during advanced abdominal pregnancy [7].

Ultrasoundography remains the main test for diagnosis, and The Royal College of Obstetricians and Gynecologists has recommended the use of the sonographic criteria described by Gerli et al for diagnosis: absence of an intrauterine gestational sac, absence of dilatation of both uterine tubes and of complex ovarian masses, a gestational sac surrounded by intestinal loops and separated from them by the peritoneum, and wide mobility similar to flotation of the sac, particularly evident with pressure from the transvaginal transducer toward the posterior fundus. [8]. However, due to the complexity and rarity of the condition, these criteria are not always present and clear even to an experienced echographer, and it is essential to perform an additional imaging exam to elucidate the case. MRI has proven to be a great help, as it provides an easier diagnosis and helps to localize and identify the relationship between the placenta and adjacent organ and tissues. The location of the placenta can help to decide whether to continue with the pregnancy and to develop a relatively safe and reasonable treatment option and surgical planning [9].

Open surgery has been the main means of treatment for advanced abdominal pregnancies due to better control of the risk of bleeding related to placental extraction, justified only if the placenta is easily removed with low risk of bleeding. It is believed that for placental delivery to regions rich in vessels and with low mobility (pelvic ligaments, the region of the iliac cases, liver, and spleen), surgery should be meticulous to avoid separation of the placenta, which should be left in its implantation site to be resorbed spontaneously postoperatively whenever possible [10]. The use of medications to assist the absorption process has been studied, the most prominent being methotrexate, but its use remains controversial because it involves a high risk of infection due to accelerated necrosis. Moreover,
in cases where the placenta remains, we should be alert for the appearance of postoperative complications such as intestinal obstruction, infection, hemorrhage, anemia, and fistula, among others. These complications can worsen the maternal prognosis, with a mortality rate above 18% [11].

Conclusions

Abdominal pregnancy is a rare type of ectopic pregnancy and is a life-threatening situation. Although this is not a frequent situation in obstetrics, it requires attention and care by specialists so that everyone is prepared to act in the best possible way. Its biggest challenge is to make an early diagnosis, since most cases go unnoticed through the ultrasound performed in the first trimester, and when symptomatic, they do not present themselves in a specific way. In our case, we were lucky because, although our patient had her first ultrasound done in an external service and late, she did it with an experienced physician and member of our clinical staff, which ensured, besides the diagnosis, an immediate referral to the obstetric center for a better evaluation and investigation of the case. It was a great challenge because, although it is a reference hospital for high-risk pregnancy, most of the obstetrics team had never had experience with a similar case, which required many team members to seek information and ensure the best care for this patient. For this, a multi-specialized team, involving, besides obstetrics, the radiology sector, surgical oncology, vascular surgery, urological surgery, anesthesiologists, transfusion agency, adult ICU, and neonatologist, was involved, since the fetus was alive and viable according to the gestational age. Unfortunately, from the fetal point of view, we did not have the best desired outcome; however, from the maternal point of view, although our first plan was for conservative treatment with metrotexate, since the patient did not have any living child, in view of the severity of the condition, the surgical complexity, and the unforeseen events during the transopetarean section, we consider that we had the best possible outcome.

Acknowledgements

We thank Dr. José Roberto Festugatto, the teams of the Obstetric Center and Surgical Center of the Hospital Geral, as well as the neonatal ICU team for the care of the newborn and the adult ICU team for the immediate postoperative care.

Department and Institution Where Work Was Done

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Declaration of Figures’ Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

References:

1. Huang K, Song L, Wang L, Gao Z, et al. Advanced abdominal pregnancy: An increasingly challenging clinical concern for obstetricians. Int J Clin Exp Pathol. 2014;7(9):5461-72
2. Singh Y, Singh SK, Ganguly M, et al. Secondary abdominal pregnancy. Med J Armed Forces India. 2016;72(2):186-88
3. Haji A, Touni D, Laakom O, et al. Early primary abdominal pregnancy: Diagnosis and management. A case report. Int J Surg Case Rep. 2020;73:303-6
4. Zhang J, Li F, Sheng Q. Full-term abdominal pregnancy: A case report and review of the literature. Gynecol Obstet Invest. 2008;65(2):139-41
5. Gurjar R. Full-term live secondary abdominal pregnancy: a rare case report. J Obstet Gynaecol India. 2019;69(Suppl. 1):36-39
6. Dahab AA, Aburass R, Shawkat W, et al. Full-term extrauterine abdominal pregnancy: A case report. J Med Case Rep. 2011;5:531
7. Siati A, Berrada T, Baidada A, Kharbach A. Abdominal pregnancy with a healthy newborn: A new case. Pan Afr Med J. 2019;34:35
8. Graham MJ, Briggs K, McMullan R, Dorman G. Abdominal ectopic pregnancy with implantation on the rectum. Ulster Med J. 2020;89(2):101-2
9. Shurie S, Ogot J, Poli P, Were E. Diagnosis of abdominal pregnancy still a challenge in low resource settings: A case report on advanced abdominal pregnancy at a tertiary facility in Western Kenya. Pan Afr Med J. 2018;31:239
10. Rohilla M, Joshi B, Jain V, et al. Advanced abdominal pregnancy: A search for consensus. Review of literature along with case report. Arch Gynecol Obstet. 2018;298(1):1-8
11. Sib SR, Oueldraogo I, Sanogo M, et al. A full term abdominal pregnancy with an isthmic tubal implantation of the placenta. BMC Pregnancy Childbirth. 2018;18(1):448