Case Report

Delayed diagnosis and management of late second trimester intra-abdominal pregnancy

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Abstract:

Abdominal pregnancy resulting in lithopedion is a rare condition constituting only 0.0054% of all pregnancies. This is a case of a 48-year-old Gravida 6 Para 3 (3-0-2-2) who consulted at the emergency room for an ultrasound finding of abdominal pregnancy. The patient had previous imaging done which showed a live intrauterine pregnancy until her fourth ultrasound, showing fetal death *in utero*. After several months without passage of the products of conception, a repeat ultrasound showed an abdominal pregnancy. Diagnosis of abdominal pregnancies may be difficult, thus it is important to utilize other imaging modalities to confirm the diagnosis. The mainstay for treatment for abdominal pregnancies is laparotomy. However, due to the possible severe hemorrhage that may arise intraoperatively, preoperative and postoperative arterial embolization of feeding vessels may be performed, as was done in the case discussed.

Keywords:

Abdominal pregnancy, arterial embolization, fetal death *in utero*, lithopedion

Introduction

Abdominal pregnancies, defined as implantation in the peritoneal cavity exclusive of ovarian, tubal, or intraligamentary implantation, are rare, with an estimated incidence of 1/402 births in developing countries and 1 in 10,000 births in industrialized countries.[1-7] Abdominal pregnancies constitute only 1.37% of ectopic pregnancies.[1] Even more infrequent are cases of lithopedion wherein an abdominal pregnancy evolves to fetal death and calcification, with an incidence of 1.5% to 2.0% of abdominal pregnancies and <0.00054% in all gestations, and only <300 cases published worldwide.[2,8] There are three classifications of lithopedion: True lithopedion (43%), wherein the fetus is calcified but not the ovular membranes; lithokeliphos (26%) wherein only the shell and ovular membrane are calcified;

and lithokelyphopedion (31%) wherein both the fetus and the membranes are calcified.[2,3]

Due to proximity to the other abdominopelvic organs, the diagnosis and management of abdominal pregnancies may be challenging. The cause of maternal morbidity and mortality is largely due to hemorrhage resulting from the detachment of the extrauterine placenta from other organs or major vessels located within the pelvic cavity.[1]

Case Report

A 48-year-old Gravida 6 Para 3 (3-0-2-2) sought consult at the emergency room of a tertiary hospital for an ultrasound showing an abdominal pregnancy. She has no known co-morbidities, and underwent dilatation and curettage for spontaneous abortion in 2014. Her first three pregnancies

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were carried to term with no fetomaternal complications while her two subsequent pregnancies were spontaneous abortions. At the time of the consult, she has had amenorrhea for 49 weeks.

History started 11 months before admission, wherein the patient experienced amenorrhea and had a positive pregnancy test result. She had her first prenatal check-up done in a lying-in center which was unremarkable. Seven months before admission, she had a transabdominal ultrasound done, which showed a single, live, intrauterine pregnancy at 21 weeks and 2 days age of gestation. Her second prenatal visit was unremarkable. Four months before admission, she complained of decreased fetal movement, prompting consult at a local hospital. Ultrasound was done which confirmed fetal death *in utero* aged at 22 weeks and 5 days by femoral length. She was advised to follow-up once with the passage of products of conception. However, 1 month had passed and there was still no passage of products of conception. Another ultrasound was done and revealed an intrauterine fetal demise, however, an ectopic abdominal pregnancy could not be ruled out. The patient was advised to consult in a tertiary hospital but was lost to follow-up. Eight days before admission, a repeat ultrasound was done which showed a macerated intra-abdominal pregnancy, prompting a consult at the emergency room for further management.

On physical examination, she had stable vital signs and essentially normal systemic findings. The abdominal examination showed a 12.0 cm × 10.0 cm irregular mass in the right periumbilical region which was firm, fixed, and tender on deep palpation. Speculum examination showed normal external genitalia, smooth, pink vagina, and a cervix with no gross lesions but was deviated to the left. Internal examination revealed a smooth and parous vagina, smooth cervix measuring 2.0 cm × 2.0 cm × 2.0 cm on palpation with the said abdominal mass seemingly attached to the corpus. The rectovaginal examination was essentially unremarkable.

The complete blood count, coagulation studies, and serum electrolytes were all within normal limits. Transvaginal and transabdominal ultrasounds were done and showed an abdominal pregnancy of about 16 weeks and 3 days age of gestation with no cardiac activity, measuring 10.5 cm [Figure 1]. The uterus was normal in size with thin endometrium. There were bilateral ovarian cysts and a left adnexal mass, to consider hematosalpinx.

On admission, abdominal X-ray was done which supported the finding of an intra-abdominal pregnancy [Figure 2]. Plain abdominal computed tomography (CT) scan and CT angiogram with

aortogram [Figures 3 and 4] were also done to identify feeding vessels into the intra-abdominal pregnancy for possible embolization, and showed an abdominal
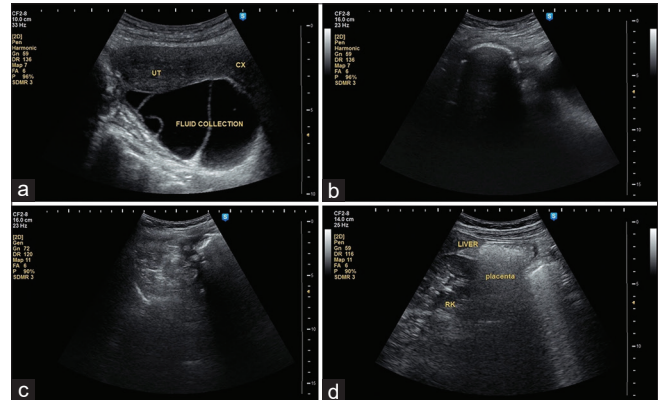

Figure 1: Transabdominal ultrasound showing an (a) empty uterus and the (b, c) extrauterine fetus and (d) placenta at 16 weeks and 3 days age of gestation described as a deformed fetus measuring 10.5 cm in length casting posterior acoustic shadows located at the right upper quadrant, inferomedial to the liver and the placenta located anterior to the fetus and below the anterior abdominal peritoneum and in close proximity to the liver, described as a heterogenous mass measuring 7.0 cm × 1.6 cm
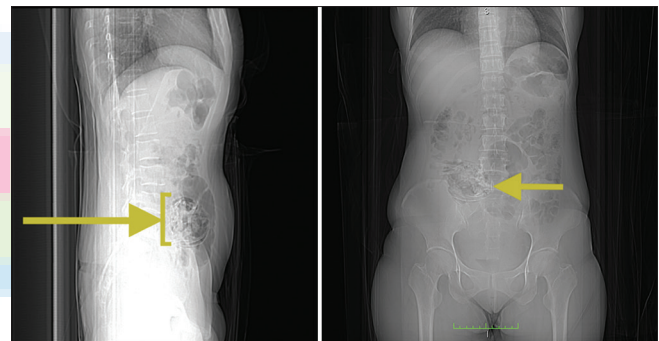

Figure 2: Sagittal and coronal views of the patient's abdominal x-ray showing the bony fetal calvarium (yellow arrows) along the right lower abdomen
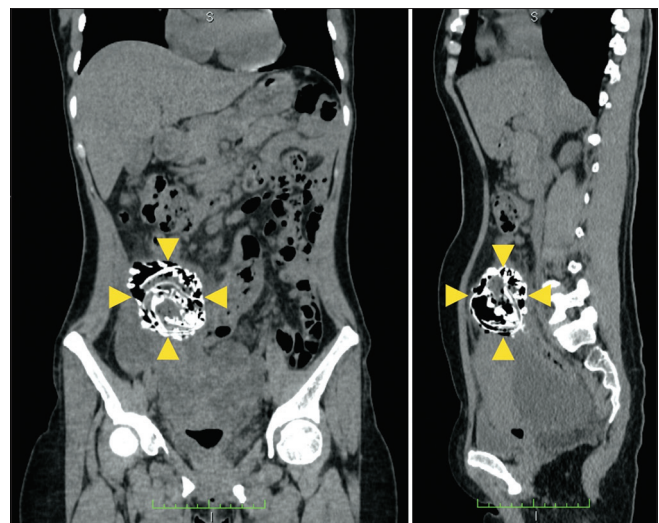

Figure 3: Coronal and sagittal views of the abdominal computed tomography scan detailing the bony fetal head (outlined by yellow arrow heads) seen in the right lower abdominal region described as a contracted fetus with disorganized anatomy

ectopic pregnancy, described as a contracted fetus with disorganized anatomy in the right lower quadrant of the abdomen. It is extrauterine in location, seen superior to the rightward and anteriorly-deviated uterine corpus. A focal region of hypodensity is noted at the left posterior aspect of the uterine fundus, with which the fetus appears to communicate, likely representing the site of rupture. On aortogram, the extrauterine fetus receives blood supply predominantly from branches of the right ovarian, right L5, and left uterine arteries.

Preoperatively, selective arterial embolization was done by the Vascular and Interventional Radiology service. The aforementioned feeding vessels were embolized using contour embolization particles with 355–500 microns mixed with iopromide 370 mg and absorbable gel sponge injection under local anesthesia. The patient tolerated the procedure well with no noted bleeding or hematoma formation.

The patient then underwent exploratory laparotomy, adhesiolysis, enterolysis, evacuation of fetal parts and products of conception, total abdominal hysterectomy with bilateral salpingectomy, colporrhaphy, and Jackson-Pratt drain insertion with the aid of General Surgery service. Intraoperatively, there was a right pelvic mass measuring 10.0 cm × 10.0 cm with fetal bones embedded in necrotic tissue [Figures 5 and 6]. No placenta was identified. Furthermore, there was a 1 cm point of rupture at the right posterofundal area [Figure 6]. The rest of the abdominopelvic organs were grossly normal. The patient tolerated the procedure well.

Postoperatively, the patient had febrile episodes and greenish output from the Jackson-Pratt drain. Work-up for possible intra-abdominal infection was done.
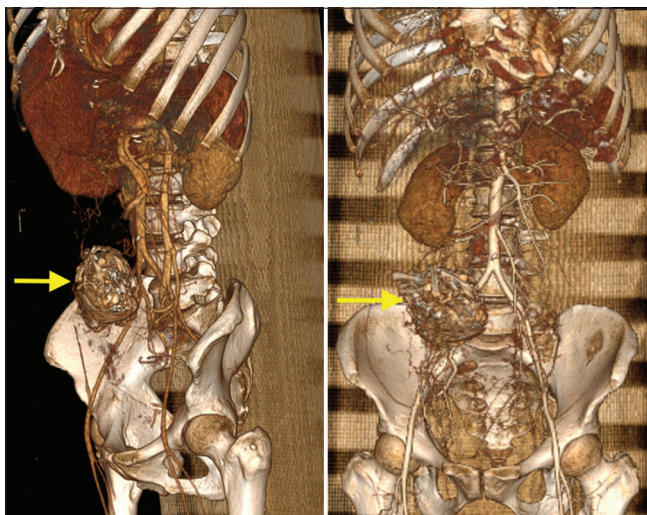
Piperacillin-tazobactam was started but eventually was shifted to Vancomycin and Meropenem. Ten days postoperatively, there was a noted 3 cm wound dehiscence on the surgical site, and the patient was started on Polymixin B. In the interim, there was the improvement of symptoms, and the patient was discharged well upon completion of antibiotic treatment.

Case Discussion

Abdominal pregnancies have a complex course and sometimes undergo calcification rather than resorption.[2] The mother's immune system treats the fetus as a foreign body and this induces calcium-rich deposition on the fetus which will eventually turn the fetal body into stone.[8] The fetal tissues undergo dehydration and calcium filtration. The prerequisites for development of a lithopedion include: (1) extra-uterine pregnancy that has escaped medical detection (2), fetal death after 3 months of pregnancy (3), fetus that has remained sterile, and (4) local conditions that favor calcium deposition.[2,3,8] The first three of these are found in the index case.

Primary abdominal pregnancies occur when the fertilized ovum implants in the abdomen.[1,4,5,7,8] However, most abdominal pregnancies are secondary to tubal rupture followed by reimplantation of the embryo into the bowel, omentum, or mesentery.[1,3,5,7] Recent literature have also discussed iatrogenic cause after *in-vitro* fertilization was done in a patient without oviducts because of previous salpingectomies.[6] Few studies have also reported abdominal pregnancy occurring after uterine rupture wherein the fetus extruded into the peritoneal cavity, which is the likely mechanism behind the index case as evidenced by the uterine rupture seen intraoperatively.[5,8]

Factors that contribute to ectopic pregnancy include congenital anomalies, endometriosis, previous pelvic infections, previous ectopic pregnancies, previous tubal and uterine surgeries, history of assisted reproductive

Figure 4: 3D reconstruction from the angiogram showing the fetal head (yellow arrow) and the vascular supply of the fetal remnants which are the branches of the right ovarian, right L5 lumbar, and left uterine arteries
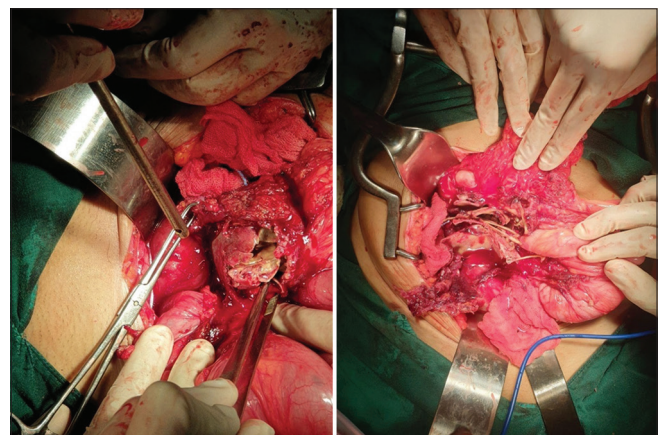

Figure 5: Intraoperative findings of fetal bones embedded in necrotic tissue
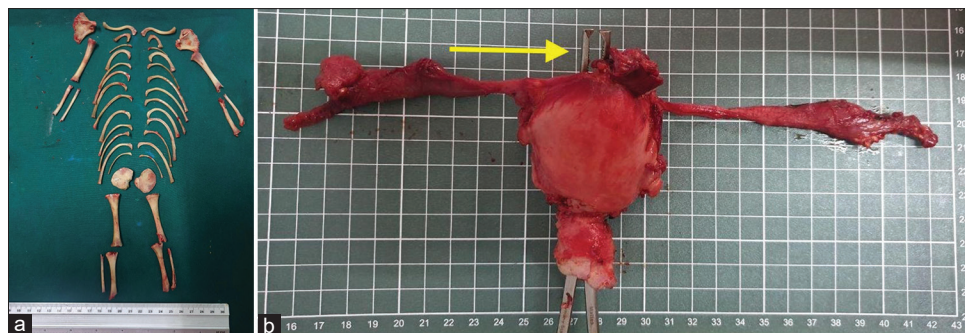
Figure 6: Intraoperative findings of (a) fetal bones and (b) uterine rupture seen at the posterofundal area

techniques, smoking, and multiparity.[3,7] Several studies have also demonstrated that the maternal age of diagnosis of lithopedion is between 23 and 100, two-thirds of which are >40.[3,8] In this case, the patient's risk factors include her two spontaneous abortions, history of dilatation and curettage, multiparity, and age of 48.

Abdominal pregnancies present with nonspecific signs and symptoms, thus rendering diagnosis difficult. The most frequent complaints include persistent abdominal or suprapubic pain (100%), bloody vaginal discharge, gastrointestinal symptoms (70%), painful fetal movements (40%), general malaise (40%), and altered bowel movements.[1-3,6-10] On physical examination, the uterus will not be palpable and the fetal lie may be persistently transverse or oblique.[4] However, most patients remain asymptomatic during large periods thus the condition may go undetected until an advanced gestational age, as in the case discussed.[2,9] Diagnosis is rarely established before surgery, therefore most are diagnosed only after presenting with some complications.[7,9]

Early diagnosis is essential because maternal complications arise as the pregnancy continues due to the involvement of large vessels and vital organs in implantation.[3,9] Maternal mortality is higher (up to 20%) in abdominal pregnancies compared to tubal and intrauterine pregnancies due to intra-abdominal bleeding which may result in anemia, infections, disseminated intravascular coagulopathy, pulmonary embolism, and fistulae formation caused by infiltration of fetal bones.[3-5,7,8] Perinatal or neonatal mortality is at 83%–95% while the prognosis for fetal survival is 11%.[7,9] Few abdominal pregnancies continue to the third trimester and to birth.[6]

Despite the variety of diagnostic imaging options, only 20%–40% of cases are diagnosed preoperatively.[9] A case review conducted in the USA found that only 11% of abdominal pregnancies were diagnosed before surgery.[9] Studdiford (1942) established three diagnostic criteria of a primary peritoneal pregnancy: (1) presence of normal tubes and ovaries, with no evidence of recent or past injury; (2) no evidence of uteroperitoneal fistula; and (3) presence of a pregnancy exclusive to the peritoneal surface and early enough in gestation to eliminate the possibility of secondary implantation after primary nidation of the tube.[10] Sonographic findings suggestive of abdominal pregnancy include: (1) absence of intrauterine gestation, (2) gestational cavity surrounded by loops of bowel and separated by peritoneum, (3) wide mobility similar to fluctuation of the sac, (4) absence of myometrial tissue between the maternal bladder and the pregnancy, (5) poor definition of the placenta, (6) oligohydramnios, and (7) unusual fetal lie.[1,4,7,9]

Based on Studdiford's criteria, the index case is a secondary abdominal pregnancy because of the uterine rupture found at the posterofundal area which may have served as the uteroperitoneal fistula from which the conceptus extruded. While the etiology behind the uterine rupture is unclear, this may have been secondary to the physical insult caused by the dilatation and curettage that the patient underwent few years before consult. Several case studies have reported similar occurrences wherein secondary abdominal pregnancies were due to the migration of the fetus through a rent in the uterus caused by previous attempts of pregnancy termination.[5]

The likelihood of misdiagnosis is also a possibility in abdominal pregnancies. The diagnostic error is estimated to be at 50%–90% in several case series, especially as the gestation develops, thus highlighting the importance of a first-trimester scan.[7,9,10] Despite the established diagnostic sonographic criteria, an advanced abdominal pregnancy may be misinterpreted as intrauterine due to the proximity of the intestines and other abdominal structures. While this is plausible for the index case, the consistent finding of intrauterine pregnancy through the different scans done by varying sonologists suggests the extrusion of the embryo through the uterine rupture rather than misdiagnosis.

In addition to ultrasound, CT and magnetic resonance imaging (MRI) may be used to establish the diagnosis and to aid in determining the involvement of other organs, in distinguishing anatomic and potential vascular connections, and in assessing the level of placental adherence, with MRI yielding better prospects.[1-6] According to Gilcha *et al.*, (2018) the most common sites of placental attachment are the uterus and adnexa (47%), bowel (30%), and the anterior and posterior pouches (8%).[6] The liver (4%), omentum (4%), and abdominal wall (4%) were less frequent sites of attachment.[6] In the case, however, no placenta was identified which may have been resorbed earlier.

Abdominal X-ray and diagnostic laparoscopy may also be utilized, as was also done for the patient [Figure 6].[3-6] Excretory urography and enema x-ray may be used to evaluate compression or other changes in the organs close to the site of implantation.[2,3] In rare cases, however, the pregnancy is diagnosed after a failed induction of labor due to the lack of myometrial response to oxytocin stimulation.[6] Several case reports have also shown diagnosis made only during cesarean section.[6]

The mainstay of management for abdominal pregnancies is surgery.[1,4,6,10] Once diagnosis is established, laparotomy with removal of the fetus should be performed immediately to prevent possible fatal hemorrhage. There is high risk for massive bleeding due to the abnormal placental attachment to extrauterine structures including large vessels.[1,6] In a review of 225 case reports, the mean blood loss was 1450 mL, with a range of 50–7500 mL, and 25% of patients requiring blood transfusion.[6]

In cases wherein the placenta is tightly adherent to the bowels and blood vessels, the placenta may be left *in situ* and is expected to resorb.[2] While this minimizes hemorrhage, this also predisposes the patient to necrosis, pelvic abscess, and wound dehiscence resulting in abdominal pain, intermittent fever, and partial bowel obstruction.[8,9] Moreover, this necessitates follow-up with β-hCG levels.[1,4,6,10] On the other hand, partial or complete removal of the placenta is preferred but may result in massive hemorrhage and shock if the vessels are not identified and ligated properly.[6,9] In the index case, however, no placenta was identified intraoperatively despite evidence of such in the previous imaging done. It is unclear why there was no placenta identified intraoperatively; however, it can be deduced that there was some form of placental involution since the pregnancy had already terminated 4 months before admission.

To prevent hemorrhage, preoperative and postoperative selective arterial embolization may be done.[1,4,9] This involves occlusion of feeding vessels using synthetic materials such as a microcatheter and absorbable gel sponge injection or coils.[1] As seen in the case, the right ovarian, right L5, and left uterine arteries were the main vascular supply of the fetal remnants and thus were embolized preoperatively to prevent severe hemorrhage intraoperatively. Data on the morbidity and mortality on use of vascular embolization are limited; however, complications include vessel puncture which may result in hematoma, pseudoaneurysm, arteriovenous fistula, dissection, intraluminal thrombosis, and embolus.[1] The use of methotrexate as adjunctive therapy is controversial. Its use may aid absorption of the retained placenta; however, it also increases the risk of infection and placental lobular necrosis which may further aggravate intraabdominal bleeding.[1,4,9]

Complications of abdominal pregnancies after a long asymptomatic course include urinary bladder perforation; rectal perforation; extrusion of fetal parts through the abdominal wall, rectum, and vagina; and intestinal obstruction.[2] These are usually attributed to the delayed diagnosis and mismanagement of the placenta.

Conclusion

Abdominal pregnancies occur either due to the primary implantation at the peritoneal cavity or secondary to the expulsion of the conceptus into the abdomen via tubal or uterine rupture. Because of its nonspecific signs and symptoms, clinical diagnosis may be challenging and a high index of suspicion is necessary given the patient's risk factors and when there is the history of insult or manipulation of the pelvic organs.

More complications arise as the gestation is prolonged, thus it is important to establish the diagnosis as early as possible. Ultrasound may not be enough to confirm the site of gestation, thus CT and MRI may be used to ascertain the site of gestation and to determine the involvement of other organs and vessels. The gold standard for the management of abdominal pregnancies is surgical management. This may be supplemented with preoperative and postoperative arterial embolization to prevent excessive hemorrhage, especially in cases wherein the placenta is adherent to vascular structures. Alternatively, the placenta may be left intraabdominal but has been documented to have negative side effects and thus is not routinely recommended.

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Conflicts of interest
There are no conflicts of interest.

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