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■ Case Report

Maternal and Foetal Outcomes in Abdominal Pregnancy: A Case Series and Review of The Literature

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Summary

Abdominal pregnancy occurs when there is implantation of the embryo in the abdominal cavity. It constitutes about 1.4% of ectopic pregnancies with an incidence ranging from 1:10,000 to 1:30,000 pregnancies. We report five cases of abdominal pregnancy with one case of fetal survival and review the existing literature. The patients presented with abdominal pain, bleeding per vagina and vaginal discharge. One was asymptomatic. All patients were haemodynamically stable except one. They eventually had surgery following ultrasound diagnosis of abdominal pregnancy. The placenta was left insitu in one of the cases. There was no maternal mortality but only one of the babies survived. Abdominal pregnancy is rare and diagnosis may be challenging, requiring a high index of suspicion. It is associated with fetal wastage. Surgical treatment is a reliable option of management, associated with good maternal outcome.

Keywords. Abdominal pregnancy, ectopic pregnancy, laparotomy

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Introduction

Abdominal pregnancy is rare and occurs when there is partial or total implantation of the embryo in the abdominal cavity. It constitutes about 1.4% of ectopic pregnancies with an incidence ranging from 1:10,000 to 1:30,000 pregnancies.[1] Risk factors include poorly treated pelvic inflammatory disease, previous history of ectopic pregnancy, infertility, tubal sterilisation, use of progesterone only contraceptive pills, tubal reconstruction, low socioeconomic status and pregnancy with an intrauterine device.[1,2] Abdominal pregnancy can either be primary or secondary. Primary abdominal pregnancy is rare and criteria for diagnosis (Studdiford's criteria) include normal tubes and ovaries, absence of uteroplacental fistula and sufficiently early diagnosis to exclude secondary implantation.[2] Secondary abdominal pregnancies originate in the tubes or ovaries and reimplant in the peritoneum. Implantation site can be anywhere in the

abdomen with the pouch of Douglas being the most common location.

Symptoms are nonspecific and abdominal pain is present in most cases. Diagnosis could be challenging. Gestational age at diagnosis is late in countries where facilities for ultrasound scan, B-human chorionic gonadotropin measurement and laparoscopy are not fully accessible.[3] It may be missed in the antenatal period and remain undiagnosed, rarely progressing to term. The maternal mortality associated with it is 90 times that of a normal pregnancy and delivery.[4] It is also associated with a high perinatal mortality. Advanced abdominal pregnancy is uncommon and guidelines for management are not clearly defined as few cases have been reported in the literature. Therefore, case reports remain important in improving diagnosis and management.

We report five cases of abdominal pregnancy, managed surgically at The University of Benin Teaching Hospital with good maternal but poor perinatal outcomes.

Case presentation

Case 1

A 30-year-old gravida 11 para 7 with three early miscarriages and 5 living children who was referred from a private hospital following ultrasound diagnosis of abdominal pregnancy with intrauterine fetal death at 42 weeks' gestation. There was no associated labour pain, liquor drainage, or bleeding per vagina. Her first contact with the referring centre was at 30 weeks' gestation with complaint of abdominal pain. Ultrasound scan done was reported by the radiologist as viable intrauterine pregnancy. There were no comorbidities. At presentation, her haemodynamic status was stable. The abdomen was distended, and fetal parts were easily palpated. Fetal heart sound was not heard. The cervical os was closed. A diagnosis of abdominal pregnancy with fetal demise was made. She was counselled on the findings and the need for surgery. Haemoglobin estimation was 9.4g/dl. Other investigations were within normal limit.

On opening the abdominal cavity, 500ml of haemoperitoneum was suctioned. There were flimsy adhesions between the membrane and gut which were easily separated. A 4.1kg macerated female fetus was extracted which had no external obvious congenital anomaly (figure 1) The placenta was attached to the right fallopian tube (figure 2), gut and omentum which was carefully separated. A right total salpingectomy was performed as the tube was adherent to the placenta. Haemostasis was subsequently secured with application of stitches and ligation of vascular pedicles. Abdominal drain was left insitu which was later removed. Blood loss at surgery was one litre and she was transfused with four units of whole blood. She was discharged after eight days and has remained stable.

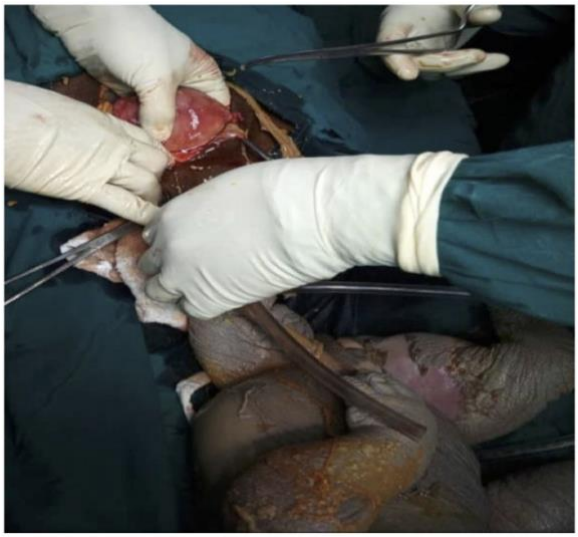

Figure 1: Dead newborn being extracted from the abdominal cavity.

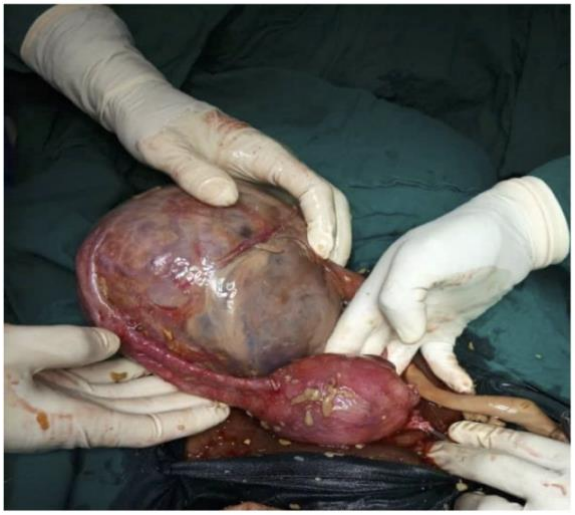

Figure 2: Appearance of the uterus with placenta attachment to the right fallopian tube

Case 2

A 31-year-old gravida 5 para 0 with four early miscarriages who was referred from a private

hospital following ultrasound diagnosis of viable abdominal pregnancy at 35 weeks' gestation. She complained of abdominal pain. Initial ultrasound scan done at 15 weeks was said to be intrauterine.

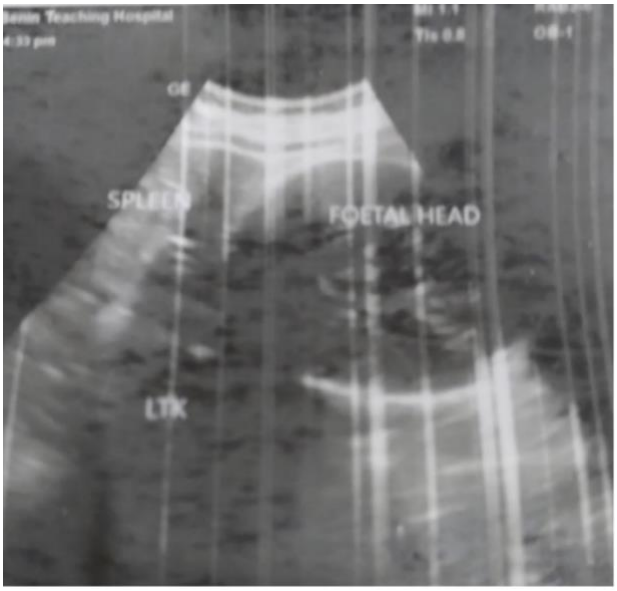

Figure 3: fetal head close to the spleen in the maternal abdomen

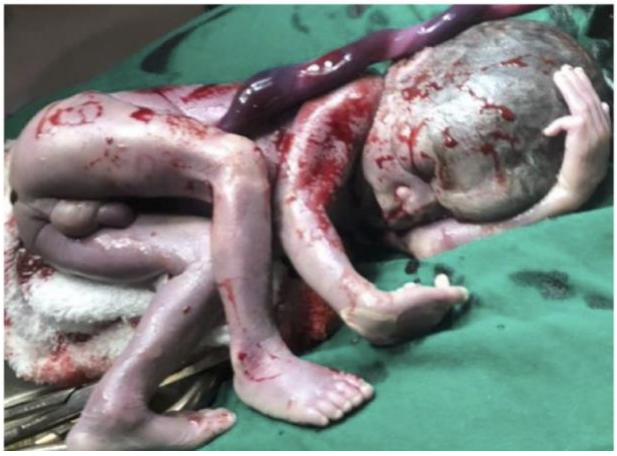

Figure 4: extracted fetus without any obvious congenital anomaly.

There were no other comorbidities. She was haemodynamically stable. The abdomen was distended, and fetal parts were easily palpated. Fetal heart sound was 140 beats/minute. Repeat ultrasound scan showed abdominal pregnancy with fetal head located close to the spleen (figure 3). A diagnosis of viable abdominal pregnancy was made. She was counselled on findings and management plan. Full blood count, urinalysis, electrolytes, urea, creatinine, and random blood glucose were within normal limit.

At surgery, a live female 2.6kg neonate (no obvious external congenital anomaly) was extracted from the abdomen. The baby later had an unexplained early neonatal death. The placenta was attached to the pelvic

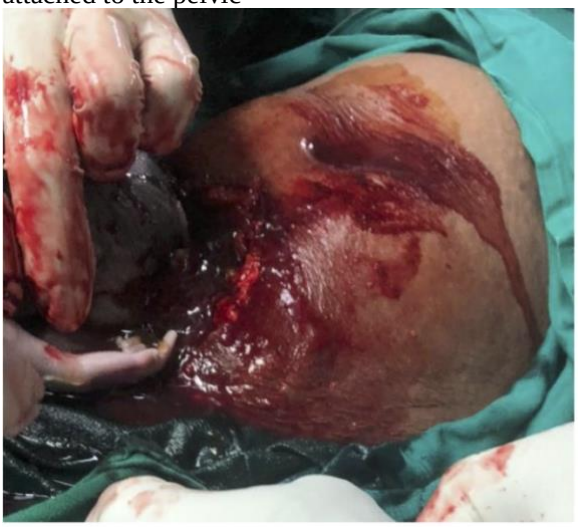

Figure 5: bulging membranes within the abdominal cavity

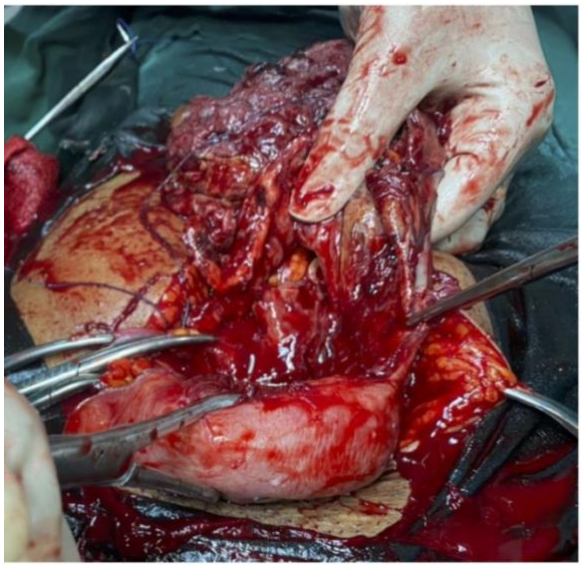

Figure 6: Careful separation of the placenta from surrounding tissues

vascular structures (bowel and omentum with highly vascular connections), hence was left insitu and patient was given methotrexate. Blood loss at surgery was 200ml. Though surgery was not haemorrhagic, abdominal drain was left insitu and removed in the postoperative period. She was

discharged home on the 11th postoperative day. She was readmitted after 8 weeks with features of sepsis and anaemia for which she was managed with antibiotics and blood transfusion. After 5 days of admission, she was discharged.

Case 3

A 26-year-old gravida 5 para 3 with 2 living children and one previous early pregnancy loss. She presented at 26 weeks' gestation with complaint of abdominal pain and slight bleeding per vagina. Her previous ultrasound scan was reported as intrauterine pregnancy. Following fetal demise, a repeat ultrasound was requested which showed abdominal pregnancy and confirmed fetal demise. There were no comorbidities. She was counseled on the diagnosis and the need for surgery. Preoperative investigations were within normal limit.

At surgery, a 0.9 kg still birth was extracted from the abdomen without any obvious external congenital anomaly (figure 4 and 5). The placenta was attached to the bowel with flimsy adhesions and was already partially separated following delivery of the baby hence was easily removed at surgery (figure 6). Blood loss at surgery was 400 ml. Abdominal drain was left insitu which remained non-functional until it was removed. She was discharged home on the 4th postoperative day and has remained stable on follow up.

Case 4

A 35-year-old Gravida 3 Para 1 with one living child and one previous early pregnancy loss who was referred to our facility from a maternity at 24 weeks gestation with complaint of vaginal discharge of 3 days duration, abdominal pain, and fever of one day duration. There was no associated bleeding per vaginam. Obstetric ultrasound scan was reported as viable intrauterine fetus with anterior placenta and severe oligohydramnios. At presentation she was febrile (Temperature 37. 7^{o}C), pale, anicteric, not dehydrated and no pedal edema. She was tachycardic (Pulse rate 118 bpm), and her blood pressure was 110/80 mmHg. The abdomen was not tender. Fetal heart was not demonstrable. On vaginal examination, there was a yellowish vaginal discharge and cervical os was closed. An impression of chorioamnionitis with fetal demise was made. She was counselled on the findings and the plan of delivery for which she consented. Full blood count done showed leucocytosis (20,000 cells/ul) with neutrophilia (81%); haematocrit was 26%. Other investigations were essentially normal. She was commenced on parenteral broad-spectrum antibiotics (ceftriaxone, genticin and metronidazole). Uterine evacuation was commenced with misoprostol for the first two days which was changed to oxytocin on the third day as the cervix did not respond. On the sixth day she had intracervical dinoprostone which also failed to achieve cervical dilatation. She was then re-evaluated and a repeat scan showed empty uterus and non- viable fetus in the peritoneal cavity. She was counseled on the diagnosis and the need for exploratory laparotomy. She had 2 units of blood transfused pre-operatively for a haematocrit of 26% bringing it to 31%. Multidisciplinary management was instituted including the general surgeons, vascular surgeons, and haematologist.

She subsequently had exploratory laparotomy with findings of haemoperitoneum (100ml), placenta attached to gut, omentum, left fimbriae and ovary, macerated stillbirth male neonate weighing 560g (no obvious external congenital anomaly), normal bulky uterus of about 10 weeks size with no fistula noted, grossly normal right tube and right ovary, estimated blood loss was 2.3 litres. The placental adhesion with guts, omentum, left tube and ovary was carefully clamped, separated and ligated. Haemostasis was secured and abdominal drain left in the Pouch of Douglas. She received five units of fresh whole blood, while abdominal drain was removed after four days. The postoperative period was unremarkable, and she was discharged home on the 8th postoperative day and has remained stable at follow up.

Case 5

A 32-year-old gravida 6 para 3 with two early miscarriages and 3 living children who was referred from a private hospital following ultrasound diagnosis of abdominal pregnancy at 31 weeks' gestation. She had no other complaint. She was HIV sero-positive. At presentation, her haemodynamic status was normal. The abdomen was mildly distended. Fetal heart sound was normal. The cervical os was closed. A diagnosis of abdominal pregnancy was made. She was counselled on the findings and need for surgery. Haemoglobin estimation was 9.2g/dl, she also had mild

hypokalaemia and other investigations were within normal limit.

At surgery, gestational sac was seen in the left broad ligament with scanty liquor and there was no communication with the uterus. A 1.2kg neonate with good Apgar scores was extracted (no obvious congenital anomaly), placenta was attached to the posterior broad ligament and was removed. Haemostasis was subsequently secured with ligation of vascular pedicles. Estimated blood loss at surgery was 650ml. Abdominal drain was left insitu and removed when it was no more active. She had a total of 4 units of blood transfused. Post-operative period was unremarkable, and she was discharged after 5 days. Baby was also managed in the neonatal intensive care unit and discharged. She has remained stable on follow up.

Discussion

Abdominal pregnancy is rare. These case series demonstrate high rate of maternal survival even in advanced cases of abdominal pregnancy and challenges encountered in its management. Diagnosis is challenging and there is no clearly defined guideline for its management. It is associated with a high incidence of fetal and neonatal mortality. The prevalence of abdominal pregnancy was 1 in 2,500 deliveries.

Mortality in abdominal pregnancy is up to 50%.[5] This was not the found in this series where there was no maternal mortality. Mortality mainly arises from haemorrhage and sepsis.[5] One major reason for the good maternal outcome was adequate preoperative preparation and multidisciplinary care which was adopted in the management of these patients. Potent antibiotics were also used to prevent infection especially when the placenta was left insitu. The clinician must always consider the risk of severe haemorrhage and possible maternal mortality if removal of the placenta is a favoured option. If this is adjudged to be a potential life-threatening procedure especially when the placenta is attached to vascular pedicles, the placenta should be left insitu as seen in this case series. Again, the high rate of fetal demise and possibly reduced placenta function may have resulted in less bleeding and reduced maternal mortality following surgery.

Only case 5 went home with a live baby who was without any congenital anomaly. Fetal wastage is a major challenge in abdominal pregnancy. The causes of foetal demise in abdominal pregnancy have not been well researched. The association of congenital anomalies with abdominal pregnancy is one of the factors causing increased incidence of perinatal death among such neonates. Fetal demise following abdominal pregnancy is as high as 80 – 100%.[5-7] Implantation in less vascular abdominopelvic organs may reduce placenta perfusion and increase the risk for fetal demise in abdominal pregnancy. Early recognition and delivery as seen in case 5 may also reduce perinatal wastage associated with abdominal pregnancy.

The clinical features of abdominal pregnancy are non-specific, and patients may be asymptomatic. Abdominal pain as seen in four of our patients, is the most constant finding in abdominal pregnancy and this could be severe in some cases.[2,3,8] Other features include nausea, vomiting, malaise, fetal malpresentation, fetal demise, bleeding per vagina and easily palpable fetal parts.[8-11] Cases 1,2,3 and 4 had abdominal pain while case 3 also reported bleeding per vagina. The cervix may also not show any signs of labour in women with abdominal pregnancy. It was therefore not surprising that although one of our patients was a grand multipara at term, her cervix remained unfavourable. Infact there have been cases where induction of labour was conducted for women with abdominal pregnancy who subsequently had surgery for failed induction of labour.[11] Case 4 had induction of labour for intrauterine fetal death but was re-evaluated following poor response to oxytocics. Intrauterine growth restriction may also be found in advanced abdominal pregnancy.[1]

The diagnosis of abdominal pregnancy could be challenging. Criteria for primary abdominal pregnancy include an early diagnosis which was not done in any of the cases presented. In one of the cases, the placenta was partly in the fallopian tube, and this should be appropriately described as secondary abdominal pregnancy. It could be missed by both obstetric palpations and ultrasonography. There is therefore need for a high index of suspicion.

In all cases, the initial ultrasound scans were not done in our facility. None had an early booking ultrasound in our facility. The prerequisite skill for ultrasonography may have been lacking at the peripheral centers from where they were referred. When ultrasonography is inconclusive, laparoscopy becomes an option in the diagnosis of ectopic/abdominal pregnancy.[2] Some authors have advocated magnetic resonance imaging as the most acceptable method of diagnosing abdominal

pregnancy.[12] Magnetic resonance imaging also identifies the relationship between the placenta and the great vessels or other vital structures which is important during surgery.[6] This may however not be readily available and accessible in a low resource setting.

There are no universally accepted guidelines for the management of abdominal pregnancy. Expectant management is an option but it carries a high risk of life threatening haemorrhage.[6] This may be considered in asymptomatic women with advanced abdominal pregnancy so as to improve fetal survival.[13] Laparotomy is generally recommended following a diagnosis of abdominal pregnancy.[3] The use of laparoscopy in the management of abdominal pregnancy has also been described.[14] During laparotomy the baby is extracted with or without the placenta. Options for placenta management including the use of methotrexate are yet to be standardized.[11] Extraction of the placenta is justified if it is easily achievable otherwise the placenta is left in-situ and spontaneous reabsorption is awaited.[4] The decision to leave the placenta is also influenced by the skill of the surgeon as haemorrhage from placental separation may be torrential.[9] Leaving the placenta in-situ may be a focus for sepsis and maternal death. Techniques used to control bleeding are compression of bleeding site, ligation of vascular pedicles, use of coagulation promoting drugs and hysterectomy for cases that have attachment to the uterus.[1] In four of our cases, we extracted the placenta, and this was associated with quick maternal recovery. In case 2 where the placenta was retained, she had a more turbulent postoperative recovery period which was complicated by sepsis and anaemia. Treatment with methotrexate may have a role in this adverse outcome. Ablation of the placenta following delivery of the fetus has also been reported.[4] Other complications of abdominal pregnancy are bowel obstruction, fetal malformations and fistula formation.[3,4]

Conclusion

Abdominal pregnancy is rare with non-specific symptoms and diagnosis may be challenging, requiring a high index of suspicion. It is associated with fetal wastage. Surgical treatment is a reliable option of management, associated with good maternal outcome. Extraction of the placenta at surgery when it is safe reduces maternal morbidity. There is need to report more cases of this rare type of pregnancy so that clear guidelines for its management can be documented.

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