A CASE OF ADVANCED NON-VIABLE ABDOMINAL PREGNANCY WITH BILATERAL EQUINO-VARUS

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Abstract

Background: Abdominal pregnancy is a variety of ectopic pregnancy with intraperitoneal implantation exclusive of tubal, ovarian or intraligamentous pregnancy. Abdominal pregnancy is very rare with a frequency directly related to the frequency of ectopic gestations in the population. The worldwide incidence ranges from 1:1320 to 1:10200 births. *Objective:* To report a case of abdominal pregnancy in the South-South region of Nigeria and highlight the clinic-radiological diagnosis of this condition. *Methodology:* The medical record of the patient was reviewed then literature of the relevant subject was medline searched. *Case Report:* A 30- year old primigravida presented to the Central hospital Ughelli in the South- south Zone of Nigeria for ANC at 12weeks gestation by date and complained of recurrent paraumbilical pains of 2 months duration. Clinical examination revealed stable vital signs but the uterus was not palpable. An obstetric scan done later showed a singleton inactive fetus, extra-uterine in location. Other significant findings were oligohydramnios and an extra-uterine placenta. The G.A was 22 weeks 1day +/- 14 days . An impression of abdominal pregnancy with fetal death was made. Radiology is central in the definitive diagnosis. Ultrasonography is recommended as a rapid non-invasive method to aid in diagnosis. The classic finding is an empty uterus separate from the fetus (100% of cases diagnosed). At emergency laparatomy the findings included a dead fetus attached to the greater omentum. *Conclusion:* The diagnosis of advanced abdominal pregnancy by ultrasound is vital to avert morbidity/mortality.

Keywords: Abdominal pregnancy, ultrasonography, extra-uterine placenta and fetus, Nigeria

Introduction

Abdominal pregnancy is a variety of ectopic pregnancy with intraperitoneal implantation exclusive of tubal, ovarian or intraligamentous pregnancy.[1,2]

Abdominal pregnancy is very rare with its frequency being directly related to the frequency of ectopic gestations in the population. About 1-2% of all pregnancies are ectopic and abdominal pregnancy is much more uncommon accounting for 1-4% of all ectopic pregnancies[3]. The worldwide incidence ranges from 1:1320 to 1:10200 births.[4]

In the USA, there is an estimated 10.9 abdominal pregnancies per 100,000 live births and 9.2 per 1000 ectopic pregnancies.[2]

A report from Afikpo, Nigeria by Sunday-Adeoye *et al*[5] places the frequency at 34 per 100,000 deliveries, while from UCH Ibadan, Ayinde *et al*[6] reported an incidence

ratio of 1 in 654 deliveries accounting for 4.3% of ectopic pregnancies.

Abdominal pregnancy is classified into 2 types; Primary and Secondary abdominal pregnancy. A primary abdominal pregnancy refers to a pregnancy that implants directly in the abdominal cavity. It is a rare phenomenon with only 24 cases reported in world literature as at 2007.[4] Secondary abdominal pregnancy is more common. It occurs when a pregnancy initially occurs in the tube and

latter on tubal abortion occurs and the fetus implants in the peritoneal cavity.[4] Advanced abdominal pregnancy is defined as a pregnancy of over 20 weeks gestation with a fetus living or showing signs of having once lived and developed in the mother's abdominal cavity.[7] Advanced abdominal pregnancy is rare and accounts for 1 in 25,000 pregnancies.[8]

Risk factors for abdominal pregnancies are the same as for ectopic pregnancy and include infertility, previous pelvic infections (PID) with tubal damage, congenital anomalies, endometriosis, previous ectopic pregnancy, assisted reproductive techniques and multiparity.[1,8-10] Cocaine use has recently been determined as a risk factor for abdominal pregnancy. Audin *et al*[11] analysing the risk factors of 55 cases of abdominal pregnancy found that cocaine use correlated with a 20% rate of increase in the incidence of abdominal pregnancy compared with a 70% decrease in the "before cocaine" time period.

The diagnosis of abdominal pregnancy requires a high index of suspicion. Clinical history, physical examination, laboratory and ultrasonographic features are all non-specific[10]. While historically plain abdominal X-ray, hysterosalpingography has been used to make a diagnosis of abdominal pregnancy, sonography is currently the most effective method for diagnosing an abdominal pregnancy. Recently MRI has started to play a role in diagnosis of abdominal pregnancy as it can provide images in multiple planes without the use of ionizing radiation.[12]

While the sonographic diagnostic error is between 50-90%[4], Allibone *et al*[13] stated the major criteria for the sonographic diagnosis of abdominal pregnancy to include demonstration of a fetus in a gestational sac outside the uterus, or the depiction of an abdominal or pelvic mass identifiable as the uterus separate from the fetus; failure to see a uterine wall between the fetus and urinary bladder; recognition of a close approximation of the fetus to the maternal abdominal wall and localization of the placenta outside the confines of the uterine cavity.

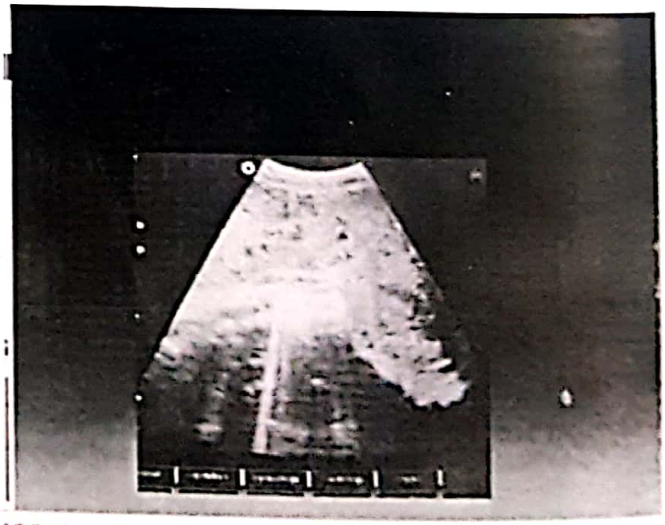
The optimum treatment of abdominal pregnancy is unknown.[9] Potential treatments consist of surgery with removal of the pregnancy via laparoscopy or laparatomy, use of methrotexate, embolization or combinations of these. The choice is largely dictated by the clinical situation.[14] If the fetus is dead, surgical intervention is generally indicated owing to the risk of infection and DIC. Some clinicians however recommend a period of observation of 3 to 8 weeks to allow atrophy of placental vessels to occur.[10] If the fetus is alive and pregnancy is less than 24weeks, immediate operative intervention is indicated. After 24 weeks there is debate as to the usefulness of careful watching (conservative management) to bring the baby to viability[10,14] i.e. 34-36 weeks.

The management of the placenta is still a matter of debate.[10] Complete removal of the placenta is advocated when the blood supply can be identified and careful ligation performed.[15] The placenta can be left insitu if there is possibility of excessive haemorrhage for natural regression with USS monitoring. This has however been associated with complications like ileus, peritonitis, abscess formation, prolonged hospital stay and fever.[10]
The introduction should be reduced to 2 pages avoiding some of the things that ought to be in the discussion.

Objective: To report a case of abdominal cyesis in the South-South region of Nigeria and highlight the clinic-radiological diagnosis of this condition.

Case Report

Mrs K.E, a 30-year-old primigravida who presented to the Central hospital Ughelli in Delta

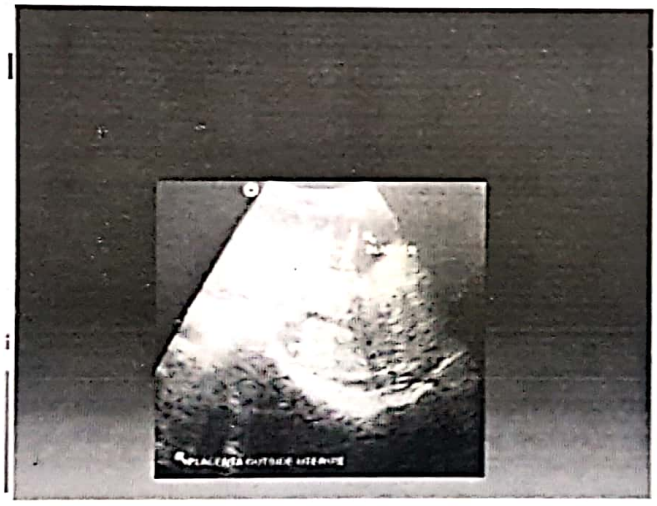


USS showing the placenta separate from the uterus

The femur length was 3.44cm which corresponded to 22 weeks 1day +/- 14 days of age *(image 2)*.

Image 2. **USS showing foetus lying transversely in abdomen. The uterus was seen to be bulky, anteverted and empty with normal myometrial echotexture.**

The endometrium showed decidual reaction *(image 3)*.



USS showing the uterus separate from the placenta and relationship to bladder

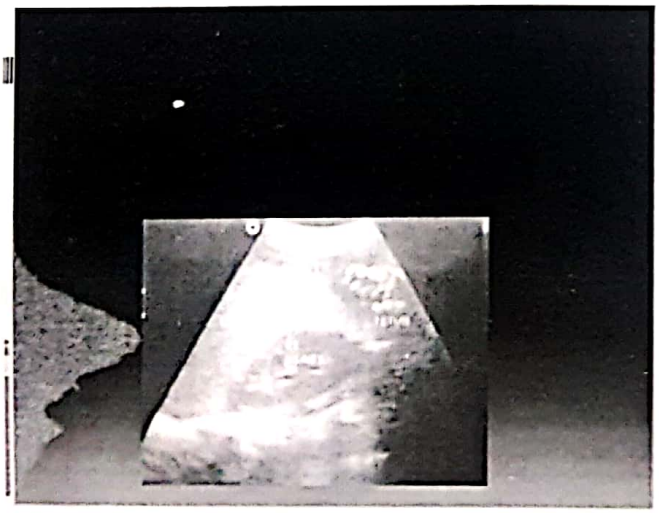


Image 4. USS showing relationship of fetus to anterior Abdominal wall, liver and kidney

state for ANC registration on the 23/4/10 and complained of recurrent paraumbilical abdominal pain of 2 months duration.

Her L.M.P was 17/1/2010 and was therefore 12 weeks by date with an E.D.D of 24/8/2010 on presentation. It was her first pregnancy after a marriage of 5 years duration in a polygamous setting. She had no history of contraceptive use in

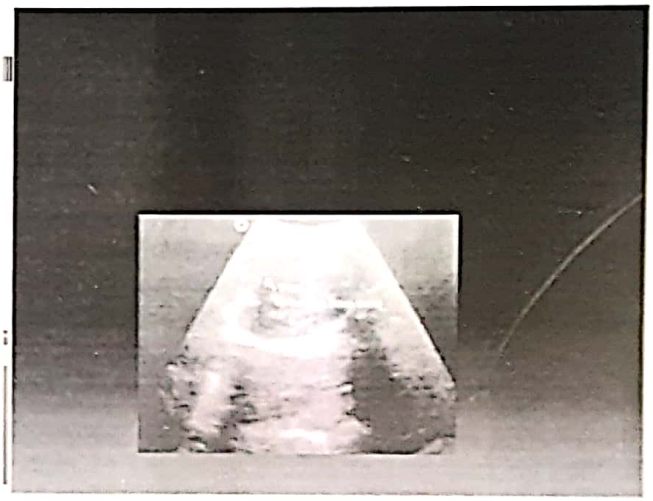


Image 5. **USS showing the fetal skull with positive spalding's sign**

the past or any history of previous operation. There was history of intermittent lower abdominal pain and vaginal discharge in the past. However there was no history of assisted reproductive technique usage.

On booking her general condition was satisfactory. Her vital signs were stable but the uterus was not palpable.

Laboratory investigations showed normal PCV (36%), HB (12mg/dl) and urinalysis. She was then placed on routine drugs.

She returned for her ANC visit 6 weeks latter and a request for a routine obstetric scan was made. On 12/7/2010 she had an obstetric scan done which showed a singleton inactive fetus lying transversely in the upper abdomen outside of the uterus in close approximation to the maternal anterior abdominal wall. The head was to the maternal left with no cardiac pulsation noted. There was marked oligohydramnios with the placenta lying outside the uterus inferior to the fetus *(image 1)*.

An impression of abdominal pregnancy with fetal death was made. Patient was immediately admitted at the Central hospital Ughelli as a result of the ultrasonographic diagnosis.

She had an emergency laparatomy on the 14/7/2010 and the findings included a dead fetus attached to the greater omentum(images 4 and 5). The placenta was noted to be outside of the uterus in an inferior position to the fetus and attached to the right broad ligament.

The fetus showed signs of maceration with a talipes equino-varus deformity of both feet. The fetus was extracted from the abdominal cavity and the placenta removed intact with haemostasis being secured. Her estimated blood loss was 500mls and she had only 1 pint of intraoprative blood transfusion.

The post-operative condition was satisfactory. She made good recovery and was discharged on the 10th day post surgery after stitches were removed.

Discussion

The occurrence of this case of abdominal pregnancy in Ughelli is not unexpected. While relatively rare, the incidence of abdominal pregnancy appears to be increasing in developing countries.[16] In a review of 163 cases reported in world literature from 13 countries since 1946, Nunyalulendho *et al*,[7] determined that one advanced abdominal pregnancy occurred for every 8099 hospital deliveries. They also noted that the incidence was 19% higher in non-industrialized countries than in industrialized countries. They therefore concluded that a physician working in a remote district (e.g Ughelli) with an active maternity service should expect to encounter several cases of advanced abdominal pregnancy during his or her working lifetime.

While the risk factors for abdominal pregnancy are the same as that for ectopic pregnancy, the higher incidence in developing countries may be as a result of the high frequency of PID in these areas.[10] It is instructive to note that our patient had a history of infertility and possible recurrent PID in the past.

The early diagnosis of abdominal pregnancy which is made based on history, physical examination and imaging is frequently difficult.[7] Clinical findings are extremely variable and pre-operative diagnosis is missed in a high number of cases.[18]

Common clinical presentations include persistent abdominal pains as was seen in this case report, being the commonest presenting symptom. Other clinical features include painful fetal movements, weight loss, vaginal bleeding, easily felt fetal parts, uneffaced cervix, abnormal fetal positions, failed induction and low haemoglobin. An elevated maternal serum alpha fetoprotein level has been associated with abdominal pregnancy particularly those with more extensive visceral implantation.[16] Our patient did not have her alpha fetoprotein levels checked as the test is not available in Ughelli.

Sonography is recommended as a rapid non-invasive method to aid in diagnosis. Stanley *et al*[18] in an evaluation of 20 proven cases of abdominal pregnancy to determine the frequency of specific abnormalities associated with this condition found identification of an empty uterus separate from the fetus as the most frequent and reliable sonographic finding. This was seen in 90% of the patients. In most cases the abdominal pregnancy was cephalad to the uterus with downward displacement and compression of the uterus and bladder. The second most frequent finding was an extrauterine placenta (75%). This was less frequent because the placenta was poorly visualized or not seen in 25% of cases. Other findings detected by Stanley *et al* included oligohydriaminos (45%), identification of fetal parts in close proximity to the maternal anterior abdominal wall (25%), failure to identify myometrium between fetal head and maternal bladder (15%), failure to identify myometrium between placenta and maternal bladder (15%), abnormal lie (25%), the presence of maternal bowel gas blocking visualization of the fetus (25%) and presence of maternal peritoneal fluid (ominous sonographic sign 10%). The final sonographic sign was the appearance of a pseudo "placenta praevia" It is instructive to note that many of these findings were present in our patient viz- (1) uterus and fetus separate (2) ectopic placenta (3) oligohydriamnious (4) abnormal lie (5) fetal parts in close proximity to maternal anterior abdominal wall (6) failure to visualize myometrium between placenta and bladder wall.

While the treatment of abdominal pregnancy depends on the clinical situation, our patient had immediate laparatomy as the fetus was already dead and complete removal of the placenta at its attachments did not envisage severe haemorrhage. Abdominal pregnancy is associated with a high maternal mortality with reported rates of 0.5-18%. The major cause of this being massive haemorrhage, which may occur during pregnancy, surgery or in the post-operative period. Other causes of maternal morbidity/mortality are infection, anaemia, DIC, pulmonary embolism and the formation of a fistula between the amniotic sac and intestine caused by penetration of fetal bone[10] It is also associated with a high perinatal mortality rate of 80-95%.[4] A report from the USA puts the maternal mortality rate at 5.1% per 1000 cases[2] while in Tanzania the perinatal mortality rate was 87.5% with a maternal mortality rate of 6.4%.[17] Ayinde *et al* reported perinatal and maternal mortality rates of 92.9% and 7.1% respectively from UCH Ibadan.[6]

Babies of abdominal pregnancy often have birth defects due to compression in the absence of the amniotic fluid buffer. The rate of malformation and deformation is estimated to be about 21%.[20] Typical deformations are facial and cranial asymmetries and joint abnormalities and the most common malformations are limb defects and central nervous system malformations.[20]

Our patient had a talipes equino-varus deformity. The sonographic diagnosis of abdominal pregnancy is extremely difficult. Atrash *et al*[2] in their analyses of 5221 abdominal pregnancies in the USA found only 1 of 9 women who reached the hospital alive had an accurate pre-operative diagnosis. Sunday-Adeoye *et al*[5] in their analyses of 20 cases in Afikpo found 50% rate of missed diagnosis while Ayinde *et al*[6] analyses of 14 cases over a 10 year period in UCH Ibadan found that the pre-operative diagnosis was possible only in half of the cases.

Conclusion

The pre-operative diagnosis of advanced abdominal pregnancy by ultrasound as in our case is unique and this presentation serves to increase the awareness and knowledge of this ominous and sometimes devastating problem. There is also the need to do pelvic/obstetric scans as early as possible.

References

1. Cotter A, Izquierdo L, Heredia F. Abdominal pregnancy. Sonoworld 2002 - 10-22-11. Retrieved 10/9/2010
2. Atrash H, Friede A, Hogue C. Abdominal pregnancy in the United States; Frequency and Mortality. Obstret Gynecol 1987; 69: 333-7
3. Isah A, Ahmed Y, Nwobodo E, Ekele B. Abdominal pregnancy with a full term live fetus: Case report. Annals of African med 2008;7(4): 198-9
4. Farhut R, Rooh M. Full term abdominal pregnancy. Prof med J 2006;13(2): 330-52
5. Sunday-Adeoye I, Twomey D, Egwatu E, Okonta P. A 30 year review of advanced abdominal pregnancy at Mater Misericordiae hospital Afikpo, South-eastern Nigeria 1976-2006. Arch Gynecol Obstret 2009. Online edition. Retrieved 12/9/2010
6. Ayinde O, Aimakhu C, Adeyanju O, Omigbodun A. Abdominal pregnancy at UCH ;A 10 year review. African J of Reprod Health 2005; 9(1): 123-7
7. Nunyalulendho N, Einterz E. Advanced abdominal pregnancy: Case report and review of 163 cases reported since 1946. Rural and remote health (online) 2008: 1087 Available from http:/www.rrh.org.au. retrieved 13/9/2010
8. Gupta P, Sehgal A, Huria A, Mehra R. Secondary abdominal pregnancy and its associated diagnostic and operative dilemma: three case reports. J Med case reports 2009; 3: 7382
9. Molinaro T, Barnhart M. Abdominal ectopic pregnancy. Semin Reprod med 2007; 25(2): 123-30
10. Kun K, Wong P, Ho M, Tai C, Ng T. Abdominal pregnancy presenting as a missed abortion at 10 weeks gestation. Hong Kong med J. 2000; 6: 425-7
11. Audin L, Brown W, Smith D, Clark J. Cocaine use as a risk factor abdominal pregnancy. J Natl Med Assoc 1998; 90(5): 277-83

12. Cohen I, Weinreb J, Lowe T, Brown C. MR imaging of a viable full term abdominal pregnancy. AJR 1985; 145: 407-8.
13. Allibone G, Fagan C, Porter S. The sonographic features of intra-abdominal pregnancy. J clin ultrasound 1981; 9(7): 383-7.
14. Abdominal pregnancy en Wikipedia/org/wiki. Retrieved 13/9/2010.
15. Hallat J, Grove J. Abdominal pregnancy a study of 21 consecutive cases. Am J Obstret Gynecol 1985;152: 444-9.
16. Parekh V, Bhatt S, Dogras V. Abdominal pregnancy: an unusual presentation. J ultrasound med 2008;27: 679-81.
17. Mbura J, Mgaya N. Advanced abdominal pregnancy in Muhimbili medical centre Tanzania. Int J Gynecol & Obstret 1986;1: 169-76.
18. Stanley J, Horger III E, Fagan C, Andriole J, Fletscher A. Sonographic findings in abdominal pregnancy. AJR 1986; 149: 1043-6.
19. Blackwelder J, Varner M, Brown R. Sonographic findings in advanced abdominal pregnancy. AJR 1981; 137: 1259-61.
20. Stevens C. Malformations and deformations in abdominal pregnancy. Am J Med Genet 1993; 47(8): 1189-95.