

CASE REPORT

Intraligamentary Extrauterine Pregnancy Delivered at Term: A Case Report and Review of Literature

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Abstract

Intraligamentary pregnancy is extremely rare. Preoperative diagnosis is often difficult. We report a case of a 33 year old female who had an ectopic pregnancy in the right broad ligament. The pregnancy remained viable till term, but unfortunately resulted in a perinatal death prior delivery. Diagnosis was not made until laparotomy and a well formed still born was delivered. A high index of suspicion and prompt therapeutic intervention are necessary to prevent adverse fetal and or maternal outcome. (*Afr J Reprod Health* 2016; 20[1]: 104-108).

Keywords: Intraligamentary pregnancy, Full term fetus, Nigeria.

Résumé

La grossesse intraligamentaire est extrêmement rare. Le diagnostic préopératoire est souvent difficile. Nous rapportons un cas d'une femme âgée de 33 ans qui a eu une grossesse extra-utérine dans le ligament large droit. La grossesse est restée viable jusqu'à terme, mais malheureusement, a donné lieu à un décès périnatal avant l'accouchement. Le diagnostic n'a pas été fait jusqu'à la laparotomie et un mort-né bien formé a été accouché. Un indice élevé de suspicion et une intervention thérapeutique rapide sont nécessaires pour prévenir une issue défavorable du fœtus et ou une conséquence maternelle. (*Afr J Reprod Health* 2016; 20[1]: 104-108).

Mots-clés: grossesse intraligamentaire, fœtus à plein terme, Nigeria.

Introduction

The diagnosis of intraligamentary extrauterine pregnancy is an enigma to the diagnostic acumen of the clinicians. The problem is further compounded in the environment such as ours where majority of pregnant women do not seek antenatal care and only present to the hospitals when complications have set in. Intraligamentary pregnancy is rare, it occurs once in approximately 245 ectopic pregnancies¹.

We report a case of intraligamentary pregnancy delivered at 41 weeks gestation in Nigeria with good maternal outcome and fully formed and grossly normal term fresh stillborn male baby.

Case Report

Mrs. S.A. was a 33 year old Gravida 4 para 2 with 2 children alive. She was referred from a Basic

Health Centre (BHC) to Ekiti State University Teaching Hospital in Ado-Ekiti, Ekiti State, Southwest Nigeria, on the 3rd of August, 2013 at 16.00hrs. She complained of abdominal pain and ten hours history of inability to perceive fetal movement. She had regular menstrual cycles of 4 days in a 28days cycle prior to conception. The first day of her last menstrual period was 18th of October 2012 and the estimated gestational age at presentation was 41 weeks and 2 days. She had registered for antenatal care at the referring BHC at estimated gestational age of 36weeks. Her booking parameters as stated in the referral note were within normal limits. She had her first ultrasound scanning at 37 weeks, which reported a viable intrauterine pregnancy with estimated gestational age compatible with 37 weeks gestation and breech presentation. Her antenatal period had been without complications till the time of referral.

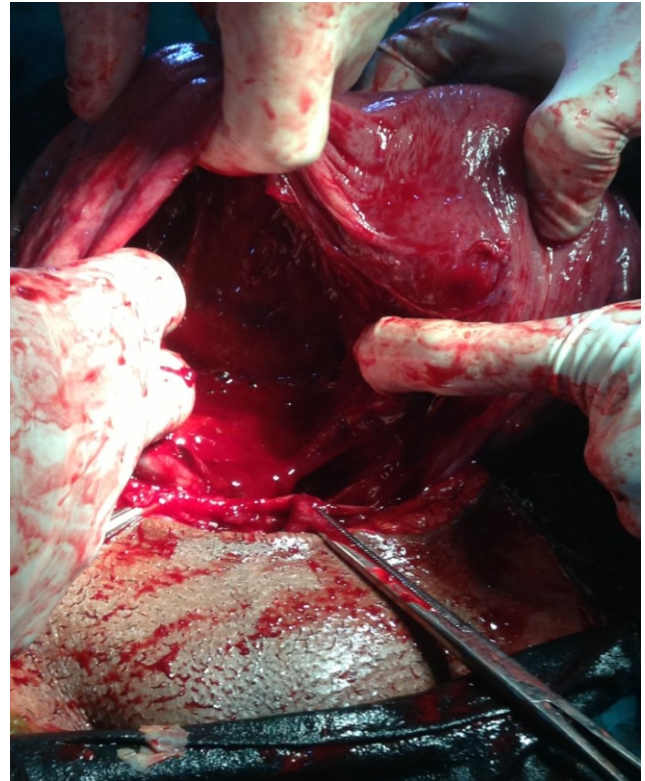
She had carried two pregnancies to term with one spontaneous abortion. Her first delivery was a live male child in 2009 by emergency caesarean section in 2009 due to intrapartum fetal distress. In 2011, she had a successful Vaginal Birth after Caesarean Section of a live female child. She was clinically stable on examination. She was not pale and her vital signs were within normal limits. The cardiovascular and respiratory systems did not reveal any abnormality. Abdominal examination reveals a healed Pfannensteil scar, symphysio-fundal height of 38cm (which was compatible with term gestation), longitudinal lie, breech presenting fetus. There were no palpable uterine contractions. Fetal heart sound was absent. She had normal vulva, a posteriorly located closed cervix about 2cm long. There was no vaginal bleeding. She had a repeat ultrasound scanning at presentation which reported an intrauterine gestation at term with breech presentation, severe oligohydramnios and no fetal cardiac activity.

We made a diagnosis of a term intrauterine fetal death with a previous caesarean section scar. Packed cell volume was 39%; blood group was O Rhesus positive, all other results were essentially normal. An attempt was made to ripen her cervix with transcervical extra-amniotic Foley's catheter followed with oxytocin induction of labour which was unsuccessful. She was then planned for abdominal delivery. Anaesthesia was induced with sodium thiopental followed by succinylcholine for endotracheal intubation. Maintenance anaesthesia was with nitrous oxide and oxygen. Intraoperatively, we found an intact uterus about 16 week's size on the infero-lateral side of the gestation.

The fetus was found within the two leaves of the right broad ligament, [Figures 1-2] surrounded by intact amniotic membranes.

The placenta was attached to the posterior aspect of the right broad ligament and the floor of the cavity in the broad ligament. The right ovary was compressed on the right pelvic side wall; the right tube was not distinctly visualized. The anterior leave of the broad ligament was incised and a fresh male still-born weighing 2.6kg was delivered [Figure3]. The right adnexum was removed with the placenta. [Figure 4] The left ovary and tube were grossly normal. Hemostasis was secured and

Figure 1: Intraoperative Photograph Showing the Space occupied by the Gestational Sac Relative to the Uterus



a rubber drain left in-situ. Blood loss was estimated at 2500ml. She had 3 units of blood transfused. Her recovery was satisfactory; post-operative packed cell volume was 30%. She was discharged from the ward on the fourth post-operative day.

A written informed consent was given by the patient to publish these records and pictures. Ethical approval was also obtained from the institution's ethics and research committee.

Discussion

Intraligamentary ectopic pregnancy is a retroperitoneal abdominal pregnancy in which the product of conception grows within the two leaves of the broad ligament. It is a rare pregnancy with an incidence of about 1 in 183,900 pregnancies². This is to the best of our ability the first report of this condition from our centre, and there have been very few reports from other centres in Nigeria^{3,4}; which underscores the rarity of intraligamentary pregnancy. Two theories had been proposed to

Figure 2: Intraoperative Photograph Showing the Space Occupied by the Gestational Sac.

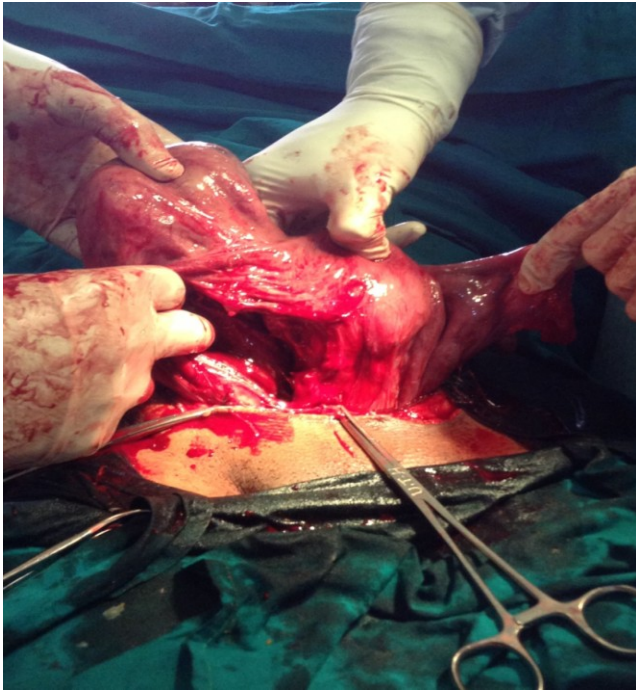


Figure 4: Post-Operative Photograph of the Placenta and Right Adnexum.

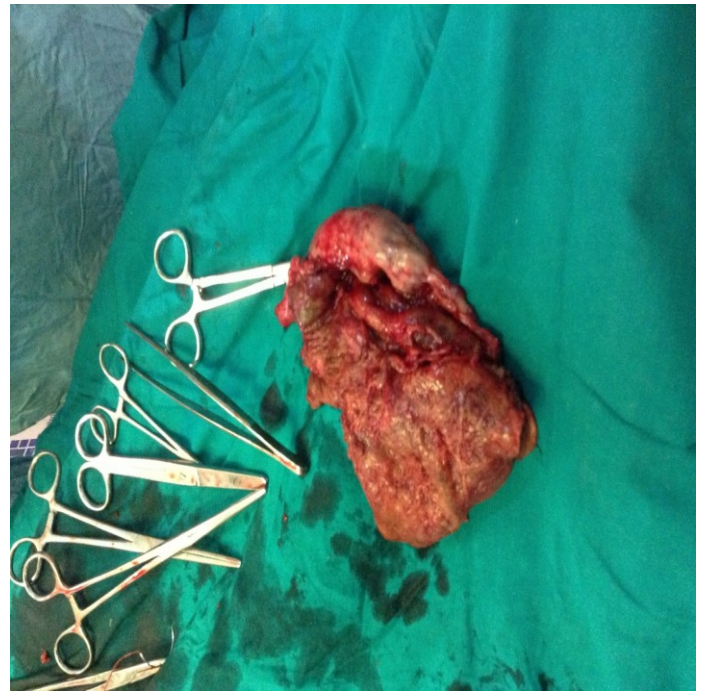


Figure 3: Photograph of a Term Fresh Male Still-Birth Delivered after a Term Interligamentary Pregnancy.



explain the pathogenesis of Intraligamentary pregnancy otherwise referred to as broad ligament ectopic pregnancy. Kobak et al⁵ explained that the majority of intraligamentary pregnancies occur as a result of the rupture of tubal ectopic pregnancies along the mesosalpingeal border with secondary escape and implantation of the fertilized ovum between the two leaves of the broad ligament. Paterson and Grant⁶ having noted cases with normal Fallopian tubes believed that intraligamentary pregnancy occurs from primary implantation of the fertilized ovum on the broad ligament with burrowing between its two leaves. Intraligamentary pregnancy can also occur if a uterine fistula develops between the endometrial cavity and retroperitoneal space⁷. The first theory probably explains the pathogenesis of the broad ligament pregnancy in our patient because the Fallopian tube on that side was not demonstrated intact. Though she had a previous caesarean section, there was no evidence of fistulous opening between the endometrial cavity and the retroperitoneal space.

The diagnosis of intraligamentary pregnancy is usually made in advanced pregnancy as occurred in our patient. This is more likely to be so in Nigeria where more than 40% of pregnant women do not seek antenatal care but only report when there is occurrence of complications^{3,4,8} as was seen in this case. Mrs. SA was only referred to our facility after failure of onset of spontaneous labour at 41 weeks gestation, for possible delivery. Early diagnosis is essential because catastrophic complications can occur due to early separation of the placenta. The associated maternal and perinatal mortality are 40% and 95% respectively^{9,10}. There was no loss of maternal life in this case, there was however perinatal death. Ultrasonography remains the main method for the diagnosis of extra uterine pregnancy⁸, however it can be disappointing in advanced pregnancy as occurred in this case.

Other radiological studies such as MRI and CT scan are helpful in the diagnosis¹¹ but are not usually available in most part of the developing world^{3,4,8}. In poor resource settings, a high index of suspicion is essential for prompt diagnosis and timely intervention to prevent catastrophic consequences. The findings of abdominal pain, abnormal lie and presentation, absent fetal heart tone, closed uneffaced cervix and the non-responsiveness of the uterus to intravenous oxytocin heightened our suspicions of this condition. The diagnosis was confirmed intraoperative with the finding of a fresh grossly normal stillborn confined between the two leaves of the broad ligament and containing the placental. This conforms to the history of ten hours history of last perception of fetal movement and ultrasound scanning findings of IUID. Bleeding from the placenta implantation site is the most life threatening complication encountered at laparotomy and the decision to remove the placenta or not can be a determining factor for survival of the woman. It is recommended to leave the placenta in situ if removal is life threatening and follow up the patient with post-operative Methotrexate therapy, hCG estimation and ultrasonography¹². Chemoembolization through lateral sacral artery has also been advocated¹³. In this case however, there was significant bleeding from the detached placenta portions that

necessitated the removal of these portions to secure haemostasis.

Conclusion

We present a case of an advanced intraligamentary pregnancy that would have led to the delivery of a healthy baby boy if the patient had presented promptly. The diagnosis of intraligamentary pregnancy requires a high index of suspicion; therefore medical practitioners should consider it a possible differential diagnosis. Ultrasonography and Magnetic resonance imaging are diagnostic tools that may help the physician in preoperative diagnosis. We also advocate early pregnancy diagnosis and community education on the importance of antenatal care in the developing countries to reduce the mortality and morbidity from ectopic pregnancy.

Conflict Of Interest

The authors declare no conflict of interest.

Contribution of Authors

Ade-Ojo, Akintayo and Aduloju all contributed to the surgery. Afolayan was the anaesthetist at the surgery. Akintayo and Ade-Ojo were the major contributors to the manuscript, Olagbuji assisted in literature search. All authors read and approved the final manuscript.

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