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Joshua Oluwafemi Aiyekomogbon, Ojah S.O., Shinkafi S.M., Agom B.

ABSTRACT

Introduction: Amniocoele is herniation of amniotic sac through a uterine myometrial defect. It commonly results from intrauterine procedures such as suction evacuation, dilatation and curettage, manual removal of placenta and other products of conception, etc. Early diagnosis is imperative as that will go a long way to reduce morbidity and mortality.

Case Report: A 35-year-old G4P3+0 women was presented to our health facility at 21st week gestation with a week history of lower abdominal pain. Obstetric ultrasound scan done at presentation revealed a fundal myometrial defect with a large amniocoele through it, resulting in apparent oligohydramnios. Her first two deliveries were uneventful, but the third was complicated by post-partum hemorrhage due to retained placenta. This necessitated instrumentation and manual removal in the theatre at a secondary Health institution, four years prior to the index pregnancy. No immediate complication was noticed after the procedure. Two weeks after this diagnosis, precisely at 23rd week gestation, she had laparotomy, aimed at reducing the herniated amniotic sac and repairing the defect. This was not possible due to the narrow neck of the defect. She was then closed back and kept on exclusive bed rest and weekly sonographic evaluation of the pregnancy. A scan done at 31st week gestation revealed ruptured herniated amniotic sac into the peritoneal cavity with fetal distress. Estimated ultrasound fetal weight at this time was 1.76 kg. She had emergency cesarean section with delivery of a live very low birth weight baby whose APGAR score was 7 and 8 at 1 and 5 minutes respectively. The baby was admitted into special care baby unit (SCBU) but died 10 hours thereafter. The myometrial defect and uterine incision were repaired in layers. She did well, hemodynamically stable and was discharged on postoperative day-7. Follow-up visits were satisfactory.

Conclusion: This case is presented to emphasize the need for training and retraining of physicians and midwives on active management of third stage of labor to forestall the observed pitfalls. It also brings to the fore the utility of gray scale and color Doppler ultrasound in antenatal diagnosis of uterine defect and placenta accreta.

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CASE REPORT

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Keywords: Amniocoele, Pregnancy, Retained placenta, 2D Ultrasound

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INTRODUCTION

Uterine perforation is a serious complication resulting from intrauterine procedure such as suction evacuation, dilatation and curettage, manual removal of retained placenta and other products of conception, insertion of intrauterine contraceptive device, hysterosalpingography, hysteroscopy and during procedures for in vitro fertilization. Invasive mole is also identified as a possible cause of uterine perforation [1–3]. Spontaneous uterine perforation from pyometra, placenta accreta, and rarely degeneration of a myoma and uterine infarction has also been documented [4, 5].

Retained placenta is one of the most common causes of postpartum hemorrhage and has an incidence of 1:100 to 1:300 births [6, 7]. Placenta adherens, incarcerated placenta and placenta accreta are the common causes of retained placenta [8]. Placenta increta is an abnormal placental implantation in which placenta villi invade into the myometrium, while in placenta percreta, placental villi penetrate through the uterine serosa or adjacent organ, usually the urinary bladder [9, 10]. Krapp et al. [6] introduced the use of color Doppler sonography in the third stage of labor to diagnose abnormal placental invasion, and differentiating it from normal placental separation. Normally, there is cessation of blood flow between placenta and myometrium immediately after birth, but in placenta accreta, a persistent blood flow from the myometrium deep into the placenta is demonstrated sonographically. This helps in differentiating placenta accreta from other causes of PPH. Using this Doppler assessment, management such as manual removal of placenta can be instituted early and by this, maternal morbidity and mortality are greatly reduced. Some biochemical markers such as elevated levels of maternal serum creatinine kinase, alpha fetoprotein, β -human chorionic gonadotropin, cell-free fetal DNA, placenta mRNA and DNA microarray are also used for the diagnosis of abnormalities of placental invasion [10–12].

Cervical dilators, curette and uterine sound are the most common instruments that cause perforation [13] and the most commonly perforated segment of the uterus is the relatively avascular fundus [14] which fits into the pattern experienced in the index case.

CASE REPORT

A 35-year-old Gravida 4, Para 3⁺⁰ (G₄P₃⁺⁰) female presented at our health facility on account of lower abdominal pain of a week duration at 21st week gestation. She had no fever, vaginal discharge or bleeding per vaginum. Her first two pregnancies were uneventful but had postpartum hemorrhage (PPH) due to retained placenta after her third delivery in 2012, i.e., four years prior to the index pregnancy. This necessitated instrumentation (curettage) after failed manual removal of retained placenta in the theatre. She was fine after the

procedure. Her blood pressure at this presentation was 120/70 mmHg and pulse rate was normal. Ultrasound of the patient done at presentation showed amniotic sac herniation through a fundal myometrial defect at 21st week of gestation (Figure 1).

The fetus was active with no sign of distress sonographically although, there was apparent oligohydramnios. She was placed on bed rest and weekly sonographic evaluation for the assessment of fetal well being. She had a laparotomy done after two weeks (at 23rd week gestational age), aimed at reducing the herniated sac and repairing the defect which also trapped the fimbrial end of the left uterine tube. Intra-operative picture is shown in Figure 2. It was difficult reducing the sac as the neck of the defect was narrow, and any attempt to force it in may lead to its rupture. Consequently, she was closed back while weekly sonographic assessment for fetal well being and exclusive bed rest were continued. The herniated sac continued to increase as the pregnancy advanced. At 30th week gestational age, the herniated sac dimension had increased to 17x18.8 cm as against the initial dimension of 6x7 cm at 21st week gestation. She noticed a sudden reduction in the size of her gravid abdomen six days after the last scan and this necessitated a repeat ultrasound scan which revealed ruptured herniated sac with features of fetal distress as shown by fetal Doppler scan (Figure 3). Estimated fetal weight was 1.76 kg at this time.

The patient had emergency cesarean section in view of the recent development, and a very low birth weight male neonate was delivered alive through a low transverse uterine incision with APGAR score of 7 and 8 at 1 and 5 minutes respectively. The child was immediately admitted into special care baby unit (SCBU) but he died ten hours thereafter. The myometrial defect (Figures 4 and Figure 5), and the low transverse incision were repaired in layers after freeing the trapped left fallopian tube. She had two units of whole blood transfused intra-operatively due to excessive blood loss. She was discharged home on postoperative day-7 after satisfactory wound healing and hemodynamic stability. The discharge hemoglobin was 11.0 g/dl and subsequent follow-up visits were satisfactory.

DISCUSSION

Uterine perforation is a defect of the uterine layers, with or without the involvement of the serosa [15]. It is a life-threatening condition for the mother and the fetus; active management is therefore imperative. It commonly results from intrauterine procedures such as suction evacuation, dilatation and curettage, manual removal of placenta to mention a few [2, 3]. We report an unusual case of myometrial defect with amniocele due to previous intrauterine procedure for removal of retained placenta four years prior to presentation. The perforation was sub-clinical and was presumably walled-off by fibrotic tissues

until the index pregnancy when the defect gave way with subsequent herniation of amniotic sac through it, resulting in severe oligohydramnios. This was diagnosed at 21st week gestation by 2D ultrasound. She was clinically stable with no sign of fetal distress initially.

Most patients with uterine perforation often present with hemorrhagic shock, abdominal pain and fetal distress. The fetus in most cases is compromised with neurological deficit because of hypoxia [16, 17]. This patient just presented with abdominal pain and the

Figure 1: Gray scale ultrasound image showing fundal myometrial defect with herniation of amniotic sac (amniocele) through the defect, which measures 1.96 cm (inverted arrow). The fetus is situated normally in the uterine cavity.

Figure 2: Picture of the first surgical exploration showing a large amniocele through a fundal uterine defect. The fimbrial end of the left fallopian tube is trapped in the defect.

Figure 3: Umbilical artery spectral waveform showing reduced end diastolic flow with elevated SD ratio (7.3) and high resistivity index (0.86), features connoting fetal distress.

Figure 4: Picture of the second surgical exploration showing a collapsed herniated amniotic sac (curved blue arrow) through a fundal uterine defect. The fimbrial end of the left fallopian tube is trapped in the defect (straight black arrow).

fetus was not in obvious distress at presentation despite the apparent oligohydramnios. No hemoperitoneum was noted clinically and radiologically, and she was hemodynamically stable with hematocrit level of 12 g/dl. The general belief is that a herniated amniotic sac into the abdominal cavity is strongly suggestive of imminent uterine rupture [18]. However, uterine rupture does not always occur immediately after sonographic

Figure 5: Picture of the second surgical exploration showing the fundal uterine myometrial defect (inverted arrow) prior to surgical closure.

detection of extrusion of the amniotic sac because a thin layer of myometrium and even uterine serosa may be around the herniated sac which may be difficult to detect sonographically [17]. Magnetic resonance imaging (MRI) scan is required to appreciate this myometrial layer and even its thickness [17]. This was not feasible in our health institution as MRI scan was not available. Serial ultrasound scan was done to closely monitor her for possible rupture instead. This was discovered at 31st week gestation but there was no hemoperitoneum despite the rupture as the uterine perforation was at the fundus which is relatively less vascular when compared to perforation at the cervical region which tends to bleed more. She only noticed a sudden reduction in the size of her gravid abdomen. This discovery, coupled with fetal distress necessitated emergency cesarean section. This was largely successful except that the neonate could not survive presumably due to birth asphyxia.

The management of amniocele depends on the gestational age and the clinical state of the mother and fetus [17, 18]. Before fetal lung maturity, two management protocols are advocated; these are surgical management and conservative management [17]. Both may be employed in some cases as it was in the index case. Surgical intervention involves termination of pregnancy and repair of the uterine wall defect, while conservative (expectant) management involves close monitoring, serial ultrasound evaluation for fetal well being and when available, serial MRI scan to evaluate surrounding myometrial thickness for possibility of rupture, and

exclusive bed rest. The index patient benefitted from both protocols. Her initial surgical management was not aimed at terminating the pregnancy but to preserve it. It was aimed at reducing the sac and repairing the defect antenatally. When that failed, conservative management was instituted. This was however abruptly terminated by sudden rupture of the herniated amniotic sac with early signs of fetal distress at 31st week gestation.

This study is aimed at preventing uterine perforation when managing third stage of labor or when conducting other intrauterine procedures for any reason, as that was the genesis of the maternal morbidity and child mortality observed in the index case. Several reports have suggested increased perforation rates by junior trainees in USA, Singapore and Nigeria [12, 19, 20]. Five-fold increase rate of perforation has been reported by junior staff in USA [12] while 82.5% of perforations were caused by junior staff in Singapore [19]. Some of these cases are even performed by untrained personnel such as Nurses, Midwives, and community health extension workers (CHEW) in Nigeria [20]. Early identification and diagnosis of uterine perforation and subsequent management will significantly reduce morbidity, long-term sequelae and even mortality [21].

CONCLUSION

Amniotic sac herniation through a myometrial defect, a sequelae of intrauterine procedures such as manual removal of placenta, dilatation and curettage and suction evacuation is globally rare but common in developing world as untrained or poorly trained personnel engage in such procedures with negative consequences as observed in the index case. The utility of gray scale and color Doppler ultrasound in antenatal diagnosis of uterine defect and placenta accreta is re-echoed.

Author Contributions

Joshua Oluwafemi Aiyekomogbon – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Ojah S.O. – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published
Shinkafi S.M. – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published
Agom B. – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor of Submission

The corresponding author is the guarantor of submission.

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None

Conflict of Interest

Authors declare no conflict of interest.

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