

Research paper

Undiagnosed Uterus Didelphys in a Term Pregnancy with Adverse Fetal Outcome: A Case Report.

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ABSTRACT

Background: Uterus Didelphys "double uterus" is a congenital malformation that results from complete failure of fusion of the ipislateral paramesonephric ducts during embryonic development. Ideally, diagnosis should be made before pregnancy and labor to prevent adverse outcomes.

Case report: Miss OE was a 32-year-old Gravida2 Para0^{+1} social mother. She was referred to Enugu State University Teaching Hospital (ESUTH) at 41weeks+ 6 days gestation for prolonged term premature rupture of membranes (PROM) and intrauterine fetal death**.** A longitudinal vaginal septum and two vaginas were later found during a critical review for failed induction of labor, and uterus Didelphys was confirmed at an emergency caesarean delivery. A macerated 3.2 kg male baby was delivered from the gravid uterus. Postoperative recovery of the mother was uneventful.

Conclusion: Clinicians should have high index of suspicion of uterine anomaly when assessing cases of dysfunctional labor to avoid delayed diagnosis and the associated adverse outcome.

Keywords: Uterus Didelphys, PROM, Failed Induction, Caesarean Delivery, Stillbirth.

Background

Uterus Didelphys is a congenital malformation of the Mullerian duct that occurs because of complete failure of fusion of the ispilateral ducts. It results in the developments of a longitudinal vaginal septum, two vaginas, and two hemi-uteri each with its separate ovary and fallopian tube. The incidence of this anomaly is 1 in 3000 [1]. It remains a challenge to the obstetricians especially when it is undiagnosed before onset of labor. Patients are usually asymptomatic, but the anomaly may be associated with dysmenorrhoea, dyspareunia, infertility, spontaneous abortion, preterm labor, fetal malpresentation, intrauterine growth restriction, PROM, renal agenesis, decreased live births and caesarean delivery [1-6].

Diagnosis is usually initiated by the findings of a longitudinal vaginal septum and two vaginal openings during a vaginal examination. A 3-D transvaginal sonography is an excellent non-invasive method of investigation [6]. Others methods of investigations include sonohysterography, hysterosalpingography, hysterolaparoscopy and pelvic magnetic resonant imaging. The incidence of caesarean delivery in uterus Didelphys in pregnancy may be as high as 82% [2]. Several good pregnancy outcomes including vaginal deliveries, twin and triplet pregnancies have, however, been reported [7-11]. The aim of this case report is to make clinicians to have high index of suspicion of uterine anomaly when investigating cases of dysmenorrhea, dyspareunia, infertility, spontaneous abortion, preterm labor, fetal malpresentation, intrauterine growth restriction, PROM, and renal agenesis. Early diagnosis, meticulous follow up can avert most of these complications

We present a case report of an undiagnosed uterus Didelphys in term pregnancy that was complicated with prolonged PROM, intrauterine fetal death, failed induction of labor, and an emergency caesarean delivery to buttress the need for early diagnosis, close monitoring in pregnancy and labor to avert adverse outcomes.

Case report

Miss OE was 32-year-old Gravida2 Para0^{+1} office attendant. She was referred to ESUTH, Enugu on 11/11/2015 for prolonged rupture of fetal membranes, and intrauterine fetal death. She attained menarche at 15 years and had a regular 28-days cycle with 4 days normal menstrual bleeding. There was neither history of infertility nor dysparenuia. She had medical abortion of a confirmed-seven-week unwanted pregnancy in 2013 without complications.

She booked for antenatal care in the index pregnancy in the referral hospital at GA of 26weeks. She complied with her antenatal appointments, investigations and drugs. There was no history of threatened abortion or preterm contractions. She received two tetanus toxoid injections and anti-malarial prophylaxis at the 24th and 30th week's gestation respectively. The pregnancy was uneventful until 41 weeks plus 6 days when she had PROM. She presented to the referral hospital where she was observed for 24 hours, had two vaginal examinations, and was later discharged without augmentation of labor. About thirty hours later she represented to the same hospital with mild uterine contractions, and was then referred.

There were mild uterine contractions at presentation. She was not pale. The pulse rate was 110 beats per minute, and blood pressure was 120/80mmHg. Her respiratory rate was 18 cycles per minute. The symphysiso-fundal height corresponded to 39weeks intrauterine pregnancy, the fetal lie was longitudinal

with 5/5 cephalic presentation. The fetal heart sounds were not heard. Initial vaginal examinations and an abdominal ultrasound missed the diagnosis of uterus Didelphys. She had intravenous Ceftriaxone 1g 12 hourly, and metronidazole 500mg 8hrly for 48 hours. Vaginal misoprostol 50 micro grammes were inserted 6 hourly x 4 without significant cervical changes. A diagnosis of failed induction was made and patient was booked for an emergency caesarean delivery. Her bedside clotting time was 5 minutes before the surgery. A critical review of the patient before surgery showed a longitudinal vaginal septum, two vaginal openings, and uterus Didelphys was confirmed at surgery as shown in figures 1 and 2. (Figure1), (Figure 2).

There were a gravid right uterus and a non gravid left uterus measuring 11cm X 6cm occupying the pelvis. Each uterus has its separate ovary and fallopian tube. A macerated male baby that weighed 3.2kg was delivered. Estimated blood loss was 1litre. She had 2 units of blood intra-operative. The postoperative recovery was uneventful. She was discharge on the 7th postoperative day after counseling her on family planning and how to manage her subsequent pregnancies. She opted for interval CuT380A intrauterine device at six week postnatal visit.

Discussion

Uterus Didelphys is rare and sometimes not even diagnosed. It occurs in 0.1% -0.5% of healthy fertile population [12]. Heinonen PK (2000) [3], evaluated the long-term clinical consequences, and reproductive performances of 49 women with uterus Didelphys that were followed up to 6.3 years. He found obstructed hemi vagina in 9 (18%) with 8 (16.3%) having ipsilateral renal agenesis. Five (13%) had primary infertility. Thirty four out of 36 (94%) women who wanted to conceive became pregnant, 21% had miscarriage while 2% were ectopic pregnancy. The fetal survival rate was 75%, prematurity 24%, fetal growth retardation 11%, perinatal mortality 5.3%, and caesarean delivery rate 84%. Pregnancy was located in the right uterus in 76% cases. Miss OE did not experience most of these complications associated with uterus Didelphys. Many of the patients with uterus Didelphys have normal sex lives, pregnancies, and deliveries [11]. Miss OE pregnancy was in the right uterus, and remained uneventful until she had prolonged term PROM, intrauterine fetal death, failed induction of labor and an emergency caesarean delivery. Her uterus Didelphys was undiagnosed until she was critically reviewed for failed induction. She missed the meticulous prenatal care that was advocated by Heine on PK

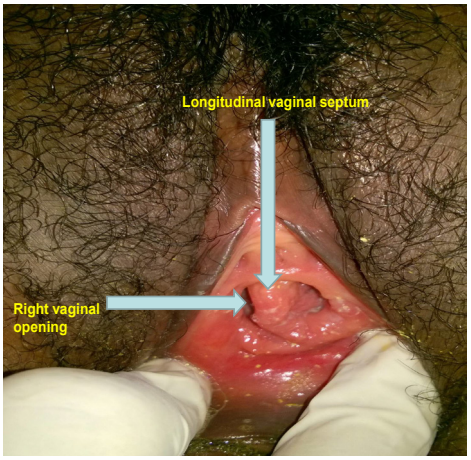

Figure1 Longitudinal vaginal septum and two vaginal openings

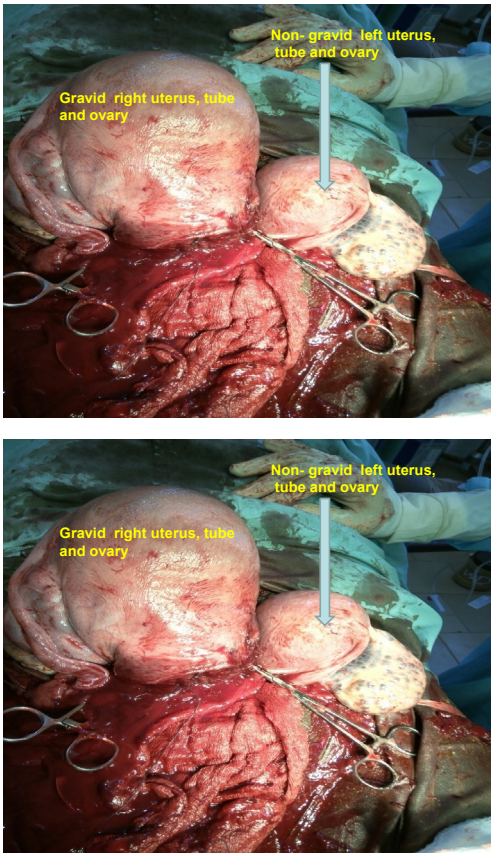

Figure 2 Two separate hemi-uteri each with a fallopian tube and an ovary.

(2000) [3] that would have prevented the adverse outcomes that were associated with this anomaly. Early diagnosis and prompt operative delivery would have prevented the fetal death in Miss OE. She had primary postpartum hemorrhage (>1 liter) due to uterine agony at surgery, and was transfused 2 units of blood intra-operative. CuT380A was not inserted during the emergency caesarean delivery to allow adequate treatment of uterine infection. Previous studies including this study were mostly case reports, and the results cannot be generalized on the general population. Only Heinonen PK (2000) [3], was able to follow 49 cases up to 6.3 years. Recent advances in diagnostic techniques, and availability of meticulous medical services and treatments for the associated complications that favor good outcomes depict the international clinical relevance of early diagnosis of the subject. None availability of such modern diagnostic technique in most developing countries like Nigeria may be the cause of the delay in establishing diagnosis, and thus delayed prompt interventions in Miss OE that could have averted the adverse fetal outcome. The directions for further studies should include universal availability of diagnostic techniques like 3 D ultrasound with vaginal probes so that population study can be undertaken, uterine anomalies identified, and protocol for the management such anomalies established.

Conclusion

Clinicians should have high index of suspicion of uterine anomaly to make early diagnosis of uterus Didelphys. Pregnancy in a uterus Didelphys deserves early diagnosis of the anomaly, and meticulous care in pregnancy and delivery to avert the associated adverse outcomes.

REFERENCES

1. Grimbizis GF, Camus M, Tarlatzis BC, Bontis JN, Dervoey P (2001) Clinical implications of uterine malformation and hysterscopic treatment results. Hum Reprod Update 7: 161-174.
2. Heinonen P K (1984), "Uterus didelphys: a report of 26 cases," European Journal of Obstetrics & Gynecology and Reproductive Biology, vol. 17, no. 5, pp. 345–350.
3. Heinonen PK (2000), "Clinical implications of the didelphic uterus: long-term follow-up of 49 cases," European Journal of Obstetrics & Gynecology and Reproductive Biology, vol. 91, no. 2, pp. 183–190.
4. Raga F, Bauset C, Remohi J, Bonilla-Musoles F, Simon C, and Pellicer A,(1997) "Reproductive impact of congenital Mullerian anomalies," Human Reproduction, vol. 12, no. 10, pp. 2277–2281.
5. Acien P (1993), "Reproductive performance of women with uterine malformations," Human Reproduction, vol. 8, no. 1, pp. 122–126.
6. Madureira AJ, Mariz CM, Bernardes JC, Ramos IM (2006). Case 94: Uterus Didelphys with Obstructing Hemivaginal Septum and Ipsilateral Renal Agenesis. Radiology 239: 602-606.
7. C. Magudapathi (2012). "Uterus didelphys with longitudinal vaginal septum: normal deliver—case report," Journal of Clinical Case Reports, vol. 2, article 13.
8. R. Garg, A. Kwatra, and V. Bangal, "A rare case of uterus didelphys with full term pregnancy in each horn," Pravara Medical Review, vol. 2, no. 4, pp. 22–24, 2010.
9. Mashiach S, Ben-Rafael Z, Dor J, Serr DM (1981) Triplet Pregnancy in Uterus Didelphys with Delivery Interval of 72 days. Obstet Gynecol 58: 519-521.
10. Magudapathi C (2012), Uterus Didelphys with Longitudinal Vaginal Septum: Normal Delivery. Journal of Clinical Case Reports**.** 2:13 http://dx.doi.org/10.4172/2165-7920.1000194.
11. Mohd Subhail, Hina Khan, Sofia Suhail (2010). Uterus Didelphys having Single Pregnancy in her right horn: A Case Report. Journal of Chinese Clinical Medicine Volume 5 (1): 46-49.
12. Green LK, Harris RE (1976).Uterine anomalies; frequency of diagnosis, and obstetric complications, 47 (4):427-428.

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