UTERUS DIDELPHYS WITH TERM PREGNANCY DIAGNOSED IN LABOR AS A CAUSE OF DYSTOCIA: CASE REPORT

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ABSTRACT

Uterus didelphys is a rare mullerian anomally associated with fertility and obstetrical complications. Term pregnancy in uterus didelphys with fetal survival is seldom reported. We report here a rare case of intrapartum diagnosed uterine didelphys associated with dystocia that gave birth to a viable baby by cesarean section.

KEYWORDS: uterus didelphys, term pregnancy, labor, dystocia

(The Ethiopian Journal of Reproductive Health; 2-20; 12;1: 55-59)

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INTRODUCTION

Uterus didelphys (double uterus) is a developmental abnormality that results from the failure of fusion of the Mullerian ducts leading to separate uterine cavities and 2 cervices [1]. It is a rare anomaly accounting for 5% of all uterine anomalies[1,2,3].

The clinical presentation is often asymptomatic and diagnosis is usually made for the 1st time during evaluation for reproductive failure or during pregnancy/ delivery[2]. It is associated with complications including dysmenorrhoea, recurrent pregnancy loss, preterm labor, cervical incompetence, malpresentation, and high cesarean section[3,5].

Thorough vaginal examination and confirmatory imaging studies including ultrasonography and MRI are important in diagnosing uterus didelphys. Treatment is surgical and reserved only for those with recurrent pregnancy loss1,6. Term pregnancy in uterus didelphys with fetal survival is seldom reported. We report here a case of uterine didelphys with a term pregnancy in the right hemi-uterus diagnosed in labor as a cause of dystocia that gave birth to an alive baby by cesarean section.

CASE PRESENTATION

A 30 years old Ethiopian primigravid lady presented to the labor ward of Karamara Hospital with pushing down pain of 20 hours and leakage of fluid per vaginum of 28 hrs. She was appreciating fetal kick. There were no urinary complaints. She didn't know her LMP. She was unbooked but had smooth pregnancy course.

She was married for 5 years and did not conceive for four years before the current pregnancy. She didn't seek medical help for her failure to conceive. Her menstrual history was unremarkable. She reported no medical illnesses.

On admission physical assessment, vital signs were stable. The uterus was 36wks sized, markedly dextrorotated. Fetus in longitudinal lie and cephalic presentation. She had two uterine contractions in 10minutes. Fetal heart beat was 146; cervix was 5cm dilated; vertex presentation, membrane ruptured.

Laboratory test results were: haematocrit: 33%, Blood group/RH, B posiitive, Random Blood Sugar = 96gm/ dl; serologic tests for syphilis, HIV and Hepatitis B were negative.

After 4hours of admission, the parturient was evaluated by an obstetrician for arrest of cervical dilatation despite adequate contractions. Speculum and digital vaginal examinations revealed two vaginal orifices with longitudinal vaginal septum extending to the vaginal outlet with two distinct cervical orifices. Right cervix was 5cm dilated.

Ultrasound was done with a clinical suspicion of mullerian anomaly (double uterus) and it revealed a singleton viable intrauterine pregnancy of 37+2 wks, cephalic presentation, reassuring biophysical profile, fundal placenta and estimated fetal weight 2.6kg normal fetal morphology. There was pear shaped, well outlined, hypoechoic mass measuring 10cm x 8cm x 3.4 cm which looks to be attached with the left lateral boarder of the pregnant uterus. Intra-abdominal viscera were normal looking. The US diagnosis was uterus didelphys with pregnancy in right hemi-uterus with possible differential diagnosis of non communicating rudimentary horn.

The patient was counselled on the possible diagnosis of soft tissue dystocia secondary to uterus didelphyis and informed consent was obtained for caesarean delivery. Under spinal anaesthesia Pfannenstiel incision was made. The intraoperative findings were: an intact, term sized gravid uterus with well formed lower uterine segment; an enlarged left sided 2nd uterus with no communication with the gravid uterus on the right side (Figure 1).

* The non-pregnant left hemi uterus is located adjacent

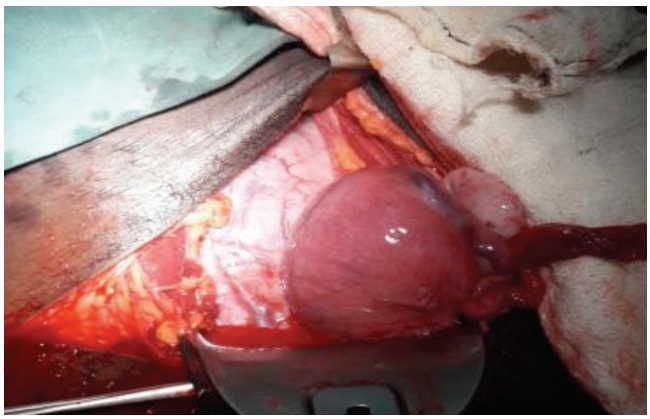

Figure 1: the anatomic relationship of the two hemi-uteri after opening the abdomen

to the lower uterine segment of the gravid uterus on the right. Note that the left ovary and fallopian tube attached with the left hemi-uterus and distended lower

uterine segment of the right hemi-uterus

A lower uterine segment transverse incision was made on the right hemi-uterus to deliver an alive neonate of weight 2.8kg with APGAR scores of 7 and 9 in the 1st and 5th minutes respectively. After repair of uterine wound, further examination showed an apparently two isthmi felt merging with the body of each of the two uteri and bilaterally normal looking tubes and ovaries each attached with the ipsilateral hemi-uterus (Figure 2).

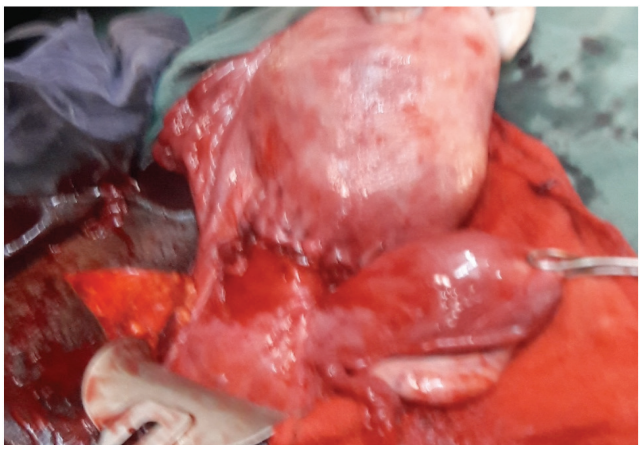

Figure 2: The appearance of the two uteri after repair of the uterine incision

* note single ovary and fallopian tube is attached with each of the ipsilateral hemi-uterus

Manual exploration of the abdominal cavity revealed that both kidneys were found at their normal location. Both mother neonate had smooth post operative course and were discharged on the 3rd post operative day. The mother was appointed for further work up and information was given for possible preconceptional visit before future pregnancy.

DISCUSSION

The uterus is derived from the paired mullerian ducts. Formation and differentiation of the mullerian ducts into the female reproductive tract depend on the completion of 3 phases of development: organogenesis, fusion, and septal resorption[7]. The process in which the lower segment of the paired mullerian ducts fuses to form the uterus, cervix, and upper vagina is termed lateral fusion. Failure of lateral fusion results in bicornuate or didelphys uterus7. Each hemiuterus is associated with 1 fallopian tube and ovary.

The reported Incidence of uterus didelphys varies from 0.03% to 0.5%1,3,8. Longitudinal septum is observed in 75% of the patients9 which was also evident in our case. Although not seen in our case, 20% of the patients with uterus didelphys have associated urinary tract anomalies [3, 10].

Most Patients with uterus didelphys are asymptomatic with the vast majority being recognized during pregnancy or as a result of recurrent pregnancy loss[4,5]. According to Jones & Jones, 1/3rd of patients with double uterus had reproductive problems[6]. The usual presenting symptoms in non pregnant women are dysmenorrhoea, dyspareunia, and infertility[4,5,10].

Pregnancy in a uterus didelphys is uncommon; the incidence varies from 1 in 1500 to 1 in 142000 pregnancies worldwide[10]. But pregnancies reaching viability and better postnatal survival were more common in uterus didelphys compared to other fusion anomalies of the uterus (bicornuate, septate or arcuate)[1,2,3]. It is however associated with obstetric complications particularly in the 3rd trimester. Pooled analyses of obstetric complications associated with Uterus didelphys reported spontaneous abortion in 32%, preterm birth in 28%. Fetal survival was in the range of 41 – 64%[1,6]. Other complications reported were cervical incompetence, prelabor rupture of membranes, Intrauterine growth restriction[3,5,7]. Poor reproductive outcome is presumed to be due to diminished uterine volume and decreased blood flow to each hemiuterus[7].

Labor and delivery complications (soft tissue dystocia) associated with uterus didelphys were well illustrated in our case that had prolonged labor with unengaged head. This could be explained by the soft tissue resistance imposed by the longitudinal septum on fetal descent and the mechanical effect of the nongravid uterus located near the pelvic brim adjacent to the lower uterine segment of the gravid uterus. Additionally abnormal fetal lies have been associated with uterus didelphys[5,7,10]. Hence Cesarean delivery rates in such patients are higher than the general obstetric population[7,12]. Yet some cases of successful vaginal deliveries have also been reported[12].

The diagnosis of uterus didelphys is suspected from genital examination where a longitudinal vaginal septum and/or two cervical orifices may be appreciated. But it remains a challenge especially pre-pregnancy as it usually is asymptomatic and affects young women where thorough pelvic examination is not possible or

contraindicated. Our case emphasizes the importance of meticulous pelvic examination in evaluation of women with labor abnormalities for possible soft tissue dystocia associated with mullerian anomalies. This will help in choosing the appropriate diagnostic study and make rational decision on mode of delivery.

Imaging studies including 2-D/3-D sonography, sonohysterography, hysterosalpingo -graphy, and magnetic resonant imaging are crucial in diagnosing uterus didelphys[1,7,9,12]. Differentiation among the duplication mulllerian problems is important as it has management implications. Two cervices must be documented to confirm uterus didelphys[1]. Three-dimensional (3-D) sonography is more accurate than 2-D because it provides uterine images from virtually any angle[9]. MRI has a reported accuracy of 100% and is usually used for difficult cases, as it has the added benefit of evaluating both internal and external appearance of the uterus[1,7,9]. Moreover, MRI is preferred in the evaluation of other anomalies possibly associated with uterus didelphys[9].

Early diagnosis is important to prevent and manage adverse obstetrical complications associated with uterus didelphys. These include hormonal support (progesterone supplementation) for recurrent pregnancy loss; cerclage for cervical incompetence and planned delivery (CD) in those with obstructive anomalies. A pre-pregnancy diagnosis of the anomaly could have been made in our case if she had visited a health facility for her infertility lasting for 4 years or if she attended antenatal care during her pregnancy. Nonetheless, unlike a similar report in the literature a catastrophe of fetal death and postpartum haemorrhage was averted in our case because of early diagnosis made in labor[13].

The definitive management of uterus didelphys is surgical correction (Strassmann's metroplasty). It is indicated only for those patients with sever dyspareunia and recurrent pregnancy loss[1,6].

CONCLUSIONS

Term pregnancy in a uterus didelphys with fetal survival is rarely reported. Uterus didelphys should be considered in the intraparum evaluation of women with suspected soft tissue dystocia. Early diagnosis and differentiation from other duplication mullerian anomalies are important to plan appropriate Individualized obstetric care. Timely interventions will avert adverse obstetrical outcomes.

ABBREVIATIONS

CS: Cesarean Section, MRI: Magnetic Resonance Imaging

DECLARATIONS

Ethics approval and consent to participate

Ethical approval for writing this case report was obtained from the health research ethics committee of the Regional Health Bureau.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests

The authors declare that they have no any competing interests.

Author's Contribution

Both WG and MB were directly involved in the evaluation and management of the case. MB collected clinical data and WG was responsible for the inception and write up of the case report.

Acknowledgements

We thank the OR staff of the maternity ward for taking the pictures during the CS.

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