DIFFERENT MULLERIAN DUCT ANOMALIES-DIAGNOSED INCIDENTALLY OR DURING EMERGENCY INTERVENTIONS
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ABSTRACT: INTRODUCTION: Mullerian duct fusion abnormalities result in different reproductive outcomes ranging from multiple uneventful childbirths to ruptures of rudimentary horn during pregnancy. AIMS AND OBJECTIVES: We intended to show the range of reproductive outcomes in different Mullerian duct anomalies. MATERIAL AND METHODS: We included 15 cases with fusion abnormalities of Mullerian ducts diagnosed for the first time while they presented to us, either by pelvic Ultrasound or during surgery. The cases of Unicornuate uterus with rudimentary horn, Bicornuate, Didelphys and Septate uterus were included with reproductive implications. RESULT ANALYSIS: One multipara woman with failed Medical Termination of Pregnancy (MTP) found to have a bicornuate uterus with undisturbed pregnancy in one horn on pelvic sonography. One primigravida woman with incomplete abortion was detected as uterus didelphys during Dilatation and Evacuation. A primigravida woman with Intra Uterine Fetal Death (IUFD) had repeated failed labor induction. By sonography, IUFD in rudimentary horn was diagnosed. We found bicornuate uterus, with pregnancy in one horn, in two mothers during Lower Uterine Cesarean Section (LUCS). Five women detected to have septate uterus during LUCS. Three cases presented with hemorrhagic shock. On laparotomy, ruptured rudimentary horn was detected; in two cases, excision of ruptured horn done but in other, the rupture site was repaired as the horn was communicating. We found one septate and one bicornuate uterus, during vaginal hysterectomy in multiparous ladies. CONCLUSION: Bicornuate and septate uterus had better pregnancy outcomes. Pregnancies in the rudimentary horns ended in critical situations. Pre-pregnancy pelvic ultrasound may help to avoid many catastrophes.

KEYWORDS: Mullerian anomalies, Reproductive outcomes, Rudimentary horn.

INTRODUCTION: Uterus developed from the fusion of bilateral Mullerian or Paramesonephric ducts. Any congenital anatomic abnormality of the uterus may have different clinical implications. The American Fertility Society (1998)¹ had classified the different Mullerian anomalies. Developmental abnormalities of the uterus affects the reproductive carrier of the women which may range from infertility, recurrent pregnancy loss or may not have any implication on pregnancy and child birth and the woman can have multiple normally delivered live babies. Among the anomalies of uterus, Septate uterus, Didelphic uterus, Bicornuate uterus or presence of a rudimentary horn do not affect pregnancy but may cause different complications (Wang et al, 2008)² et al some of which may even be life threatening, eg, ruptures of one horn during pregnancy. The presence of anomalous uterus could have been diagnosed by pelvic Ultrasound much before conception, the only way to avoid any emergency situations arising out of it. Soriano et al (1999)³ et al showed that demonstration of fetal uterus is possible at 19th week of intra-uterine life with high reproducibility. So, there is a vast period in a woman’s life, starting from her intra-uterine life, when a pelvic ultrasound would have been done to exclude any congenital uterine anomalies. Jayasinghe et al (2005)⁴ et al in a meta-analysis showed that the pre-rupture diagnosis of non-communicating horn of uterus is disappointingly low.
We had in our record a wide range of cases with uterine anomalies which we had diagnosed incidentally or during an emergency situation without any prior intimation about the anatomical abnormalities. Some of the cases had quite uneventful outcome while in other cases the conditions became very critical. Many a times it had become very difficult to make the diagnosis at the time of emergency because of unawareness about the defective structure of the uterus.

AIMS AND OBJECTIVES: We had compiled the cases to depict the wide varieties of reproductive outcomes in different congenitally abnormal uterus due to mullerian ducts fusion anomalies. We were intended to emphasize the life threatening situations arising in cases of previously undiagnosed rudimentary horn of uterus with pregnancy. To generate awareness about some of the major problems in reproductive period due to different uterine anomalies was our intention so that early pre-pregnancy diagnosis of this sort of anomalies would have been taken care of.

MATERIAL AND METHODS: We had included the cases of various uterine anomalies due to defective fusion of Mullerian ducts, which were diagnosed incidentally. None of the cases had the diagnosis of the uterine anomaly before they presented to us. In all the cases we had found the structural abnormality of the uterus either by pelvic Ultrasound done after some complications arising after the routine procedures or during any emergency surgical procedures. We could not take the help of three dimensional ultrasound or Magnetic Resonance Imaging (MRI) as these were not available in our institutions. We had followed the nomenclatures given by The American Fertility Society while classifying the Mullerian anomalies. The cases with arcuate uterus were excluded in this study. We did not also include the cases of hypoplasia or agenesis of uterus. The cases with Unicornuate uterus with rudimentary horn with or without communication with the unicorneate cavity, Bicornuate uterus, Didelphys uterus and Septate uterus were included in this study with their implications in respect to reproductive outcomes. Those cases that did not have any Obstetric complications in spite of the uterine anomaly were also included. The Institutional Ethical committee had approved the study.

RESULT ANALYSIS: We have 15 cases in our record. The events during which the diagnoses of abnormal uterus due to mullerian ducts fusion anomalies were done are depicted in Figure (Fig. 1) 1. The different types of Mullerian fusion anomalies we had found are shown in Fig. 2.

One multipara woman presented with signs of continuation of pregnancy two weeks after Dilatation and Evacuation (D & E) done outside for Medical Termination of Pregnancy (MTP) in her first trimester. On Ultrasonography, we could clearly found a bicornuate uterus with two uterine cavities, with the single cervical canal (Fig. 3). The pregnancy remained undisturbed in left horn, not approached by the previous surgical procedure. The MTP was completed by D & E, where two uterine cavities communicating with a single cervical canal could be identified. The woman was discharged with oral contraceptive pills.

One primigravida woman presented with signs of incomplete abortion after eight weeks of amenorrhoea. After initial resuscitation, she was put in the Operation table for D & E. On inspection, she was found to have uterus didelphys with two cervix and also a longitudinal vaginal septum (Fig 4). After D & E, she was advised to come for subsequent follow up but she did not.
One primigravida woman presented at 20 weeks of gestation with sonological diagnosis from outside as having Intra Uterine Fetal Death (IUFD). Induction of labor tried with Prostaglandin E₂ vaginal gel and repeated after 12 hours but there was no effect. Then labor induction tried with vaginal application of Misoprostol tablets (200 mg) at an interval of six hours for two alternate days without any result. Another sonography was taken which excluded abdominal pregnancy by showing IUFD. Again induction tried with Misoprostol and even with extra amniotic Ethacrydine Lactate instillation followed by intravenous Oxytocin infusion. After having failed with all the procedures, we did another meticulous sonography which diagnosed the case as unicornuate Uterus with the dead fetus in rudimentary horn (Fig. 5) and the horn which was approached so far with all the medications was empty (Fig. 6). Hysterotomy was done, the fetus was taken out followed by excision of the horn was undertaken as clinically no communication with the cervical canal could be established.

We have in our record, two cases, both primigravida, where we incidentally found bicornuate uterus, with pregnancy in one horn, during Lower Uterine Cesarean Section (LUCS). All of them were referred cases from peripheral centers and none had any pelvic Ultrasonography before or during pregnancy. Both had premature rupture of membranes and pre-term onset of labor and had to undergo LUCS due to non-progress of labor. Another five women had LUCS and during the surgical procedure, incomplete septum within the uterus was identified. Two of them had breech presentation at 36 and 35 weeks of gestation respectively; one was referred to us with uncontrolled Pregnancy Induced Hypertension and another woman had complaint of no perception of fetal movements for last 12 hours, with a history of previous two first trimester fetal losses without any investigations done to search the cause. The other three women had pre-term labor; two women had fetuses in transverse lie and another woman had fetus in oblique lie.

All the babies had good Apgar Scores and were doing well. All the women were discharged with proper counseling and oral contraceptive pills.

There were three cases in 18 – 22 weeks of gestation, who presented with features of hemorrhagic shock and acute abdomen. There was no history of trauma over the abdomen or any other specific antecedent history. One of them, a primigravida woman, had one sonological screening report of Intra uterine single-tone pregnancy, one week back prior to the presentation but there was no hint of uterine anomaly. The other two women, one primigravida and other had two previous live births, did not have any sonological evaluation before or during pregnancy. In all three cases laparotomy was decided after necessary resuscitation. On opening the abdomen, massive hemo-peritoneum was found with suspicion of abdominal pregnancy as the fetus was lying in the peritoneal cavity (vide the picture of one baby after removal from peritoneal cavity; Fig. 7) and one intact, slightly enlarged uterine horn was noted. But after meticulous inspection, the ruptured rudimentary horn of the uterus detected (Fig. 8, Fig. 9). In two cases, both the primi mothers, excision of the ruptured horn done as the horns were clinically not communicating with the cervical canal. In one case, the woman had previous two live babies; the rupture site was repaired (Fig. 10) with bilateral tubal ligation as the horn had communication with the cervical canal. All three women had a stormy post-operative period and survived after getting four to five units of blood transfusions.

We came across two cases, one septate and the other bicornuate uterus (Fig. 11) while doing vaginal hysterectomy. Both the menopausal women, at their sixth decade of lives, with a smooth Obstetric history, had five and six children respectively and all were normally delivered at home. The diagnosis of fusion anomalies of uterus was made only during vaginal hysterectomy.
DISCUSSION: We had met with a wide variety of reproductive outcomes in women having fusion anomalies of the Mullerian ducts. Wang et al (2008)\textsuperscript{2} in their study showed that septate uterus associated with obstetric complications eg, premature rupture of membranes and abnormal presentations. They also noted that in bicornuate uterus recurrent miscarriage and preterm labor is more common.

Many a time, the cases were confused with abdominal pregnancy as in the case of IUFD in our study, where there were recurrent induction failures. Same confusion also occurred in our study cases at the first look during Laparotomy for rupture of one horn. Allen (2007)\textsuperscript{5} et al reported a case of secondary abdominal pregnancy after rupture of one horn of Bicornuate uterus.

Rupture of rudimentary horn of uterus always gives rise to an extreme emergency situation. In our study we found three cases (at 18 - 22 weeks of gestation); in two nullipara women the pregnancies were in the non-communicating (rudimentary) horn and on Laparotomy, excision of the horn was done. One case, a multipara woman, the rupture site was repaired as the horn was communicating. Jayasinghe et al (2005)\textsuperscript{4} in their study showed that in 92% of cases of rudimentary horn were of non-communicating type. Giraudet et al (2006)\textsuperscript{6} commented, rupture of the rudimentary horn usually occurs in the second trimester; which corroborates with findings of our study. Kore S et al (2000)\textsuperscript{7} commented that Rupture uterus in nulliparous patients is generally associated with Mullerian anomalies. We recorded three cases of ruptured rudimentary horn of uterus, two of them were nulliparous.

Diagnosis of congenital abnormality of uterus should be done beforehand to avoid any catastrophe during gravid state, but all the time it is not an easy task. In our documentation, one woman had a pre-rupture sonography without detection of gestation in the rudimentary horn uterus and ultimately ended in a life threatening situation. The other case with IUFD in one horn also required repeated sonography to arrive at a diagnosis of bicornuate uterus. Jayasinghe et al (2005)\textsuperscript{4} in the meta analysis showed that the sensitivity of the diagnosis of rudimentary horn by sonography is only 26% and diagnosis before any clinical symptoms occurred is 14%. Tsafrir et al (2005)\textsuperscript{8} noted that rudimentary horn of the uterus can be diagnosed by sonography before rupture in the first trimester of pregnancy which could be confirmed by MRI with the help of some radiological criteria. Daskalakis G et al (2002)\textsuperscript{9} reported a case where even with a suspicion of pregnancy in rudimentary horn at seven weeks, the patient did not agreed to have termination of pregnancy and ultimately resulted in a life threatening rupture at 20 weeks.

On the other hand we recorded two cases of bicornuate uterus and five cases of septate uterus which were found during LUCS without any previous diagnosis of uterine anomaly. Abnormal fetal presentations were noted in all cases of septate uterus. All of them had different obstetrical complications including pre-term labor but they had live babies due to emergency LUCS could have been done as they were referred from periphery at proper time. Heinonen (2006)\textsuperscript{10} showed in his study that the live birth rate in women without metroplasty in septate uterus is as high as 72%. Troiano and McCarthy (2004)\textsuperscript{11} in their meta-analysis noted that in septate uterus, overall premature birth rates ranging from 9% to 33% and fetal survival rates from 10% to 75% and in bicornuate uterus, premature birth rates range from 14% to 23% and fetal survival rates, from 57% to 63%. As examples of extremely successful reproductive carriers, we recorded two cases that had multiple normally delivered live babies and diagnosed as having bicornuate uterus and septate uterus one each, during vaginal hysterectomy in their sixth decade of lives.
CONCLUSION: There is wide variation in the reproductive outcomes in women with different fusion anomalies of the Mullerian ducts. The cases with bicornuate and septate uterus had more favorable pregnancy outcomes. Pregnancies in the rudimentary horns had presented with critical, confusing and even life threatening situations. Pre-pregnancy meticulous pelvic ultrasound or more sophisticated diagnostic procedures like, three dimensional ultrasound or MRI during early pregnancy may be of immense help to avoid many catastrophes in the structurally abnormal gravid uterus.

REFERENCES:

1. American Fertility Society. The American Fertility Society classification of adnexal adhesions, distal tubal occlusion, tubal occlusion secondary to tubal ligation, tubal pregnancies, müllerian anomalies and intrauterine adhesions. Fertil Steril 1988; 49: 944-55.
2. Wang SJ, Oli M, Jina Q, Wang JL, Wei LH. Clinical analysis of 225 women with congenital uterine malformation. Zhonghua Fu Chan Ke Za Zhi. 2008; 43(7): 493-6.
3. Soriano D, Lipitz S, Seidman DS, Maymon R, Mashiach S, Achiron R. Development of the fetal uterus between 19 and 38 weeks of gestation: in-utero ultrasonographic measurements. Human Reproduction. 1999; 14(1): 215-18.
4. Jayasinghe Y, Rane A, Stalewski H, Grover S. The presentation and early diagnosis of the rudimentary uterine horn. Obstet Gynecol. 2005; 105(6): 1456-67.
5. Allen WL, Subba B, Yoong W, Fakokunde A. Chronic abdominal pregnancy following rupture from a bicornuate uterus. Arch Gynecol Obstet. 2007; 275(5): 393-5.
6. Giraudet G, Mubiayi N, Nayama N, Le Goueff F, Therby D. Uterine horn rupture at 23 weeks gestation: a case report. J Gynecol Obstet Biol Reprod (Paris). 2006; 35(8 Pt 1): 826-8.
7. Kore S, Pandole A, Akolekar R, Vaidya N, Ambiye VR. Rupture of left horn of bicornuate uterus at twenty weeks of gestation. J Postgrad Med. 2000; 46(1): 39-40.
8. Tsafrir A, Rojansky N, Sela HY, Gomori JM, Nadjari M. Rudimentary horn pregnancy: first-trimester prerupture sonographic diagnosis and confirmation by magnetic resonance imaging. J Ultrasound Med. 2005; 24(2): 219-23.
9. Daskalakis G, Pilalis A, Lykeridou K, Antsaklis A. Rupture of non-communicating rudimentary uterine horn pregnancy. Obstet Gynecol. 2002; 100(5 Pt 2): 1108-10.
10. Heinonen PK. Complete septate uterus with longitudinal vaginal septum. Fertil Steril. 2006; 85(3): 700-5.
11. Troiano RN, McCarthy SM. Mullerian duct anomalies: imaging and clinical issues. Radiology. 2004; 233(1): 19-34.
Fig. 1: Distribution of cases of according to the events during which the Uterine anomaly was diagnosed

Fig. 2: Different types of Mullerian anomalies in our study

Fig. 3: Bicornuate uterus showing fetal pole in left horn
Fig. 4: Uterus Didelphys with a longitudinal vaginal septum

Fig. 5: USG showing fetal head (IUFD) in one horn of uterus

Fig. 6: USG showing one empty horn and another horn with pregnancy (in transverse section)

Fig. 7: Picture of one fetus after retrieval from peritoneal cavity

Fig. 8: Rupture of the Rudimentary horn of uterus

Fig. 9: Close view of the rupture site of left horn
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