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Case Report

Obstetric Outcome in a Woman who had Unicornuate Uterus with Ectopic Ovary, Absent Kidney and Ureter on Right Side

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Keywords
• Absent kidney
• Ectopic ovary
• Obstetric outcome
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Abstract

Background: To report a case of Obstetric outcome in a woman who had unicornuate uterus with ectopic ovary, absent kidney and ureter on right side

Case Presentation: We report 23 year old third gravida short stature woman presented with 8 month amenorrhea and premature labour pain. Her first baby died due to septicemia and meningitis after first caesarean section done four year back due to breech presentation, oligohydramnios with premature rupture of membrane. Second time two years back she delivered a stillborn male child of 2 kg weight after exploratory laparatomy due to rupture of uterus when she admitted in same hospital for premature onset of labour and severe pain abdomen. In third time during caesarean we found incidentally that uterus was unicornuate with only left ovary and fallopian tube. After thorough search we found normal ovary and 3 to 4 cm of a fallopian tube in right pelvic wall without connection with uterus. After one month mother's whole abdominal ultrasonography was performed to detect urogenital anomalies and right kidney was absent. This was a very rare case in which woman had spontaneous pregnancy without infertility treatment and mullerian duct anomalies, ectopic ovary and absent kidney and ureter on right side with variable obstetric outcome.

Conclusion: Our case is of unicornuate uterus with spontaneous conception without infertility treatment. In previous pregnancies, her obstetric outcome was poor and maternal morbidity occurred during last one in form of ruptured uterus. Unicornuate uterus, Ectopic ovary, absent kidney and ureter on right side were incidental finding during third caesarean section. Also we highlight that neonates of women with urogenital anomalies possibly had high incidence of prematurity and infection.

INTRODUCTION

Prevalence of congenital uterine anomalies varies between 3.4 and 5.0% and Müllerian anomalies are found more often in sub fertile patients compared to fertile controls. Congenital malformations of the Müllerian system are probably caused by multifactorial polygenic and familial factors. The prevalence of a unicornuate uterus is rather low (0.3% of the whole population, 0.6% of the infertile population, 0.2% of the fertile population). However, of all Müllerain defects unicornuate uterus is found in 3 to 13% of women. Unicornuate uterus is caused by a failure of one Müllerian duct to develop (unicornuate uterus without rudimentary horn) or to migrate to its proper location. During the third month of fetal life the developing ovaries descend from a position near the kidneys to their final position in the pelvis, this descent is guided by the gubernaculum. The gubernaculum is attached to the uterus forming the utero-ovarian and round ligament. Ectopic or undescended ovaries are characterized by the attachment of the upper pole to an area above the level of the common iliac vessels. Incidence of Ovarian maldescent is reported to be 20% when the uterus is absent (Rokitansky-Küstner-Hauser syndrome) and more than 40% in cases of uni -cornuate uterus Despite the well-known association of ectopic ovaries and unicornuate uterus, ectopic ovaries are reported only sporadically, suggesting the possibility that many cases go unrecognized [1]. There is a close embryologic relationship

between the mesonephric and paramesonephric ducts, so that defects in the development of the mullerian structures can also lead to defects in renal anatomy, such as ectopic location, aberrant anatomy, or complete agenesis [2]. Approximately 40% of patients with a unicornuate uterus will have an associated urinary tract anomaly. The unicornuate uterus is an uncommon anomaly, representing only 4.4% of uterine anomalies. Collectively, unicornuate uteri are associated with relatively poor reproductive Outcome [3].

Some who have found an association between uterine anomalies and preterm birth (PTB) theorize that diminished muscle mass, particularly in a unicornuate uterus, plays an important role in the mechanism of preterm delivery [4]. Our case is of unicornuate uterus with spontaneous conception without infertility treatment. In previous pregnancies, her obstetric outcome was poor and maternal morbidity occurred during last one in form of ruptured uterus. Unicornuate uterus, Ectopic ovary, absent kidney and ureter on right side were incidental finding during third caesarean section. Also we highlight that neonates of women with urogenital anomalies possibly had high incidence of prematurity and infection.

CASE PRESENTATION

A 23 year old short stature woman with weight and height of 48 kilogram and 146 centimeter, third gravida with 8 month amenorrhea and premature labour pain was admitted in our hospital as booked case. Menstrual history was normal. There were no significant history of medical, surgical illnesses and treatment of infertility except previous caesarean sections. First caesarean section was done four year back due to breech presentation, oligohydramnios with premature rupture of membrane. Her 1.4 kg preterm female child was expired on 4th day due to septicemia and meningitis. Second time two year back she was admitted in same hospital due to premature onset of labour and severe pain abdomen. Medical record showed that she had rupture of uterus so exploratory laparatomy was done. She delivered a stillborn male child of 2 kg weight. Details regarding mullerian anomalies of uterus were not mentioned on previous discharge tickets. In present pregnancy when premature labour pain started she admitted and thoroughly investigated. Tocolytic and steroids were given. Antenatal ultrasonography revealed a single live fetus with cephalic presentation gestation of around 32-36 weeks, adequate amniotic fluid. Rest investigations were within normal range. Her elective caesarean section was decided after completion of 36 weeks. During caesarean we found uterus was unicornuate with only left ovary and fallopian tube (Figure 1). We thoroughly search the right adnexa and found normal ovary and 3 to 4 cm of a fallopian tube in right pelvic wall without connection with uterus (Figure 2). It was an incidental finding during surgery. She delivered a healthy male child of 2.2 kg with Apgar score of 7 and 8 on 1 and 5 minute. Mother's postoperative period was uneventful. On 2nd day baby was admitted in nursery with gasping respiration, peripheral cyanosis and heart rate of less than 60 per minute. Bag and mask ventilation was started, Baby was reviewed and kept on oxygen, fluid and antibiotics. All investigations were normal except septic screen was positive and blood culture sensitivity showed that baby had kleibsella sepsis and antibiotic was given for 14 days. After one month mother's

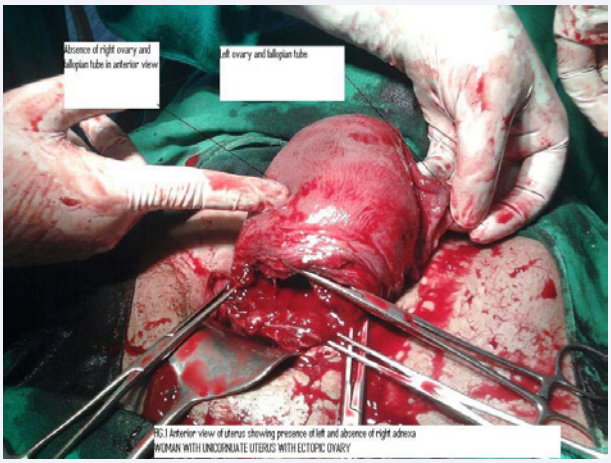

Figure 1 Anterior view of uterus showing presence of left and absence of right adnexa.

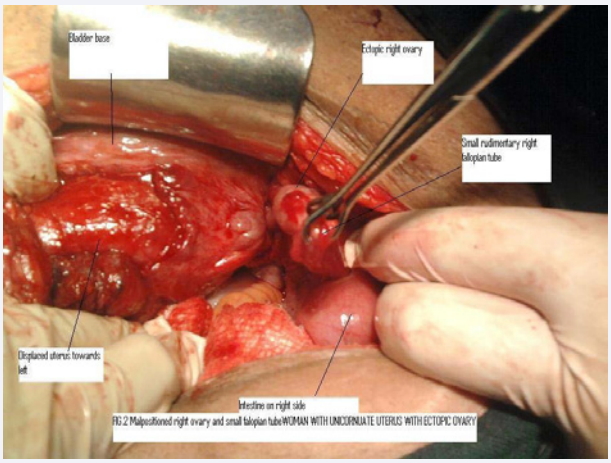

Figure 2 Malposioned right ovary and small fallopian tube.

whole abdominal ultrasonography was performed to detect urogenital anomalies and right kidney was absent. This was a very rare case in which woman had spontaneous pregnancy without infertility treatment and mullerian duct anomalies, ectopic ovary and absent kidney and ureter on right side with variable obstetric outcome.

DISCUSSION

Ectopic ovary is a rare entity associated with unicornuate uterus and urological anomalies, characterized by their attachment to an area above the level of the common iliac vessels with an incidence estimated between 1 in 29,000 and 1 in 700,000 gynecologic admissions [1]. Despite the well-known association of ectopic ovaries, urogenital anomalies and a unicornuate uterus, ectopic ovaries and urogenital anomalies are reported only sporadically, suggesting the possibility that many cases go unrecognized [2,3]. There are usually primary infertility and obstetric outcome are poor in these women [3].

A unicornuate uterus can be present alone or with a rudimentary horn or bulb on the opposite side. It is associated

with urinary and other mullerian duct anomalies. When all mullerian duct derivatives and the kidney are absent on one side, this implies failure of development of entire urogenital ridge, including the genital ridge where the ovary forms. The reproductive performance of women with unicornuate uterus is poor, with a live birth rate of only 29.2%, prematurity rate of 44%, miscarriage rate of 29%, and an ectopic pregnancy rate of 4 %.

The unicornuate uterus is an uncommon anomaly, representing only 4.4% of uterine anomalies. Collectively, unicornuate uteri are associated with relatively poor reproductive outcome. In a review of 151 women with an untreated unicornuate uterus who had a total of 260 pregnancies the mean abortion rate was 37.1%, the mean preterm delivery rate was 16.4%, the mean term delivery rate was 45.3%, and the mean live birth rate was 55.1%. However, different types of unicornuate uterus are associated with different reproductive success rates. The success rate is dependent on numerous factors that include: variations in the vascular contribution from the uterine artery and utero-ovarian artery of the contralateral side, extent of the reduction of muscular mass of a unicornuate uterus, degree of cervical competence, and presence and extent of coexistent pelvic disease such as endometriosis. In the largest series of women with a unicornuate uterus who were infertile or had recurrent pregnancy loss, the live birth rate in those with a communicating rudimentary horn was 15%, with a non communicating rudimentary horn was 28%, and with a rudimentary horn without a cavity 35%. Only one woman had a unicornuate uterus without a rudimentary horn and did not have a live birth. The highest live birth rates are observed in women with a rudimentary horn, with or without a cavity [3].

This is a rare case of embryologically congenital absence of right sided kidney and ureter, malpostioned/ ectopic right ovary that was not attached to unicornuate uterus, and ectopic ovary was found incidentally during surgery her previous obstetric outcome was poor. Only male child delivered in this pregnancy survived after septicemia. There may be chance of litigation in future so detail evaluation about urogenital anomalies should be done in women with unicornuate uterus.

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