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A CASE REPORT OF SUCCESSFUL PREGNANCY OUTCOME IN UNICORNUATE UTERUS WITH KYPHOSCOLIOSIS

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ABSTRACT Unicornuate uterus belongs to class U4 or Hemi-uterus classification according to the European society of human reproduction and embryology (ESHRE). The incidence of Hemi-uterus is 1-2 %. It is associated with fetal survival of 40%. Kyphoscoliosis is an abnormal curvature of the spine in both coronal and sagittal planes. Prevalence varies from 0.3% to 15.3%. We present a case of 27 years, unbooked primigravida with 34 weeks gestation with leaking per vagina and pain abdomen with breech presentation. She had short stature and kyphoscoliosis. During caesarean section she was found to have unicornuate uterus with absent left fallopian tube and ovary.

KEYWORDS : Unicornuate uterus, Hemi-uterus, Kyphoscoliosis

INTRODUCTION

Mullerian duct anomalies are a result of abnormal formation, fusion, or reabsorption of Mullerian ducts during fetal life. Mullerian duct anomalies are present in 5.5% in an unselected population, 12.3% in women with a history of miscarriage, 8% in infertile women, and 24.5% in women with a history of miscarriage and infertility.[1] Mullerian duct anomalies are associated with complications like growth restriction, malpresentation, preterm premature rupture of membranes, preterm birth, cesarean section, placenta previa, and placental abruption.[2]Unicornuate uterus belongs to class U4 or Hemi-uterus classification according to the European society of human reproduction and embryology (ESHRE).[3] Incidence of Hemi-uterus is 1-2 % of all uterovaginal anomalies.[4] It is associated with poor reproductive performance with fetal survival of 40%. There is a high risk of spontaneous abortions, prematurity, breech presentation, severe intrauterine growth restriction, and increased incidence of cesarean section. These anomalies have an association with skeletal deformities like scoliosis of the spine and the Klippel-Fiel anomaly.

Kyphoscoliosis is defined as an abnormal curvature of the spine in both sagittal and coronal planes. Prevalence varies from 0.3% to 15.3%.[5] Restrictive pulmonary dysfunction is the commonest reported pulmonary impairment. Women with severe spinal deformities have impaired adaptation of the thoracoabdominal architecture and are considered high-risk pregnancy. Significant effects on cardiorespiratory physiology were noted in case of a change in normal spine curvature. It also causes considerable changes in pelvic anatomy which may alter the course of labour and delivery. Primary pathology in the lumbar spine leading to kyphosis in this region has an overall effect on spinal balance and pelvic obliquity. Pregnancy in such a deformed spine imposes special challenges in anesthetic and surgical management of patients.

We report a case of a successful outcome in pregnancy with a unicornuate uterus with kyphoscoliosis.

CASE REPORT

A 27 years old, unbooked primigravida presented at 34 weeks 4 days gestational age with complaints of leaking per vagina, pain abdomen, and tightening since 2 hours. On admission she was afebrile, and her vital signs were within normal limits. She was short stature, thin built with kyphoscoliosis(Figure 1). She also had polydactyly of left lower limb(Figure 2), congenital left lower limb shortening and paralysis. Abdomen examination was suggestive of 32 weeks of pregnancy with uterus deviated more towards the right side with a longitudinal lie, breech presentation, estimated fetal weight of 1.6-1.7kgs with prominent fetal parts. Per speculum examination showed active leak of amniotic fluid. Per vaginal examination done cervix was 3/4th inch long and posterior in position, cervical os was 2 cm dilated with extended breech, absent membranes and contracted pelvis. Scan done in labor room showed 2cm amniotic fluid index and was having a non-reassuring nonstress test (NST). Her routine blood and urine investigations were done and her hemoglobin was 9.6 gm/dl. She was given antibiotics and was taken up for an emergency cesarean section. Intra-operatively she was incidentally found to have a unicornuate uterus with no rudimentary horn with absent left fallopian tube and ovary (Figure 3).

Grade 1 placenta previa was seen. There was no liquor. Delivered a preterm male baby of 1.75kg by breech extraction with good apgar. There was no post-partum hemorrhage and the uterus was closed in two layers. Her postoperative period was uneventful. Later ultrasound abdomen was done and the left kidney could not be visualized.

HRCT chest was done which showed a significant mediastinal shift to left, agenesis of left upper lobe bronchus with tubular and cystic bronchiectasis changes in left lower lobe bronchus with complete atelectasis of lower lobe with dorsolumbar kyphoscoliosis. She was discharged from the hospital on postoperative day 7. Mother and baby were healthy at discharge.

Figure 1: Kyphoscoliosis

Figure 2: Polydactyly of left lower limb

Figure 3: Unicornuate uterus

DISCUSSION

Hemi uterus is defined as the unilateral uterine development with functional uterine Hemi-cavity, with contralateral part either incompletely formed or absent. Class 4 is divided into class U4a or Hemi-uterus with a rudimentary cavity, characterized by the presence of a communicating or non-communicating functional contralateral horn, and class U4b or Hemi-uterus without rudimentary cavity characterized by the presence of nonfunctional contralateral uterine horn or by aplasia of the contralateral part.[6] It is suggested that first trimester abortion, stillbirths, and intrauterine growth restriction are a result of abnormal uterine blood flow, and second trimester abortions and preterm deliveries are thought to be a result of decreased muscle mass in the unicornuate uterus as well as cervical incompetence. They may be associated with urinary tract anomalies like renal agenesis, malrotated kidney, low lying or pelvic kidney, and need to be investigated by intravenous pyelogram. Wang et al. presented 26 cases of unicornuate uterus with rudimentary horn, four of which (15%) had become pregnant, and one of which (4%) had been presented with ipsilateral oviduct ectopic pregnancy.[7] Fox et al, suggested that the risk of adverse pregnancy outcomes was increased in patients with uterine unicornis and the rate of preterm birth was almost 50%.[8] According to American Congress of Obstetricians and Gynecologists (ACOG) current guidelines for the management of IUGR are to consider serial growth ultrasound examinations in pregnancies at risk of IUGR as in a unicornuate uterus pregnancy. These deformities can complicate pregnancy which leads to challenges for obstetric surgeons. Such patients often have skeletal anomalies like congenital scoliosis which may involve any part of the spine. Involvement of the spine may lead to a difficult approach to spinal anesthesia and cardiorespiratory consequences. The etiology of kyphoscoliosis is tuberculosis, trauma, neuromuscular disease osteochondrodysplasia, connective tissue disorders, and degenerative changes.[9] The striking feature, in this case, was that despite severe deformity with the unicornuate uterus there was no cardiorespiratory embarrassment during pregnancy. The presence of kyphosis in the lumbar region has a direct effect on the lower abdomen and pelvis. If the deformity of the spine is significant, there is an approximation of the ribcage to the iliac crest resulting in the reduction of the available room in the abdomen. As a result, there is an acute angulation of the growing uterus resulting in a pendulous abdomen. Due to distorted anatomy, cesarean section in such a severely kyphotic patient presents unique challenges. Chau and Lee reported out of the total twenty-five cesarean sections carried out in kyphotic patients, a classical incision was given in twelve cases as the lower segment was inaccessible. An incision on the fundus and even on the posterior surface of the uterus has been described for delivery of the fetus.[10] Postpartum hemorrhage may be anticipated due to atonicity of the uterus due to uterine inertia which is reported in cases of kyphotic patients due to the acute angulation of the uterus which causes the fetal axis to divert from the axis of the birth canal. Cesarean section in a severely kyphotic patient presents with unique challenges. In the event of future pregnancy, the patient should be advised of the need for strict supervision at a tertiary care center with a multidisciplinary approach and the need for preplanned elective cesarean with proper preoperative anesthetic assessment.

CONCLUSIONS

The reproductive outcome for unicornuate uterus is poor but a successful pregnancy is possible with good diagnosis and screening of antenatal cases with serial ultrasound assessment.

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