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Successful Delivery of Twins in a Woman with a Unicornuate Uterus

Abstract

The presence of a unicornuate uterus is a rare congenital condition which represents only 1-2% of uterovaginal anomalies. Previous reports have demonstrated an increase in the number of cases of primary infertility, pregnancy loss and preterm labor associated with the unicornuate uterus. Herein, we present a case in which a patient conceived following a thawed frozen embryo transfer. She had been given the option of selective reduction, because of the high risk associated with this pregnancy, but refused and her pregnancy successfully continued to 35 weeks and delivery of twins.

Key Words

Unicornuate uterus

Twin gestation

Frozen embryo transfer

Introduction

A unicornuate uterus is a rare congenital anomaly representing 1-2% of all uterovaginal anomalies [1, 2]. Poor reproductive function is frequently associated with this condition [3]. A greater incidence of primary infertility, pregnancy loss and preterm labor has been reported with congenital uterine anomalies. Unicornuate uteri, in particular, have shown the highest rate of primary infertility (15%) and the poorest fetal survival (40%) [3]. The management and outcome of a patient with a left unicornuate uterus and a twin gestation is discussed.

The unicornuate uterus is a rare diagnosis. Andrews and Jones [4] were able to find only 8 women with this diagnosis in their entire infertility and obstetric practice at Norfolk General Hospital and Johns Hopkins Hospital between 1969 and 1981. This defect is considered to represent about 1-2% of all uterovaginal anomalies [1, 2]. A review of the literature from 1959 to 1983 showed that in 119 pregnancies in 50 patients with unicornuate uteri, there was a spontaneous abortion rate of 33%, a preterm delivery rate of 29%, and a live birth rate of 66% [5], whereas another study estimated fetal wastage to be 31% [2].

The management and outcome of a pregnant patient with a unicornuate uterus and a twin gestation is described herein.

Case Report

A 29-year-old nulliparous white female presented with the complaint of primary infertility of 4-year duration. The patient who had a history of anovulation was treated with human menopausal gonadotropins (hMG) for 3 months without achieving a pregnancy. She was known to have a left unicornuate uterus from a diagnostic laparoscopy 1 year before. More recent laparoscopic findings revealed fimbrial agglutination of the left fallopian tube, congenital absence of the proximal portion of the right oviduct, and the presence of both ovaries. Hysterosalpingography showed a smaller than usual uterine cavity. Intravenous pyelogram showed medullary sponge kidneys with no other abnormalities noted.

At this point, the patient was given the option of reconstructive surgery or in vitro fertilization. She elected the latter treatment. She underwent controlled ovarian hyperstimulation with hMG following pituitary suppression with leuprolide acetate. During her first cycle, 21 oocytes were retrieved transvaginally from 27 follicles. Twelve oocytes fertilized, 5 embryos were transferred and 7 embryos were frozen (4-6 cells each) using a modification of the one-step technique [6, 7]. However, a pregnancy was not achieved at that time.

Ovulation was subsequently induced with clomiphene citrate for a frozen embryo cycle. When two 19-mm follicles were present on the

right ovary, human chorionic gonadotropin was given and 3 frozen embryos were transferred. Individual embryos suspended in the cryoprotectant 1.5 M 1,2-propanediol were aspirated into 0.25 ml freezing straws. Separated from the embryo by a 1-cm air column, the remainder of the straw was filled with 1.08 M sucrose. The straws were seeded at -6.0°C and held for 15 min in an alcohol bath, rate-controlled freezer (FTS Systems, Stoneridge, N.Y., USA). The straws were cooled at $-0.4^{\circ}\text{C}/\text{min}$ to -35.0°C and held again for 15 min before plunging into liquid nitrogen. The cryoprotectant was diluted out in one step after thawing each straw by vigorously shaking the straw to mix the sucrose with the embryo column, incubated in 37°C water for 3 min and cooling the straw for 1 minute in room temperature water. Three of the 4 thawed embryos survived and were cultured for 15 h overnight before transfer. All 3 underwent cell division prior to embryo transfer.

A twin gestation was documented by ultrasound at 6 weeks gestation. From 20 weeks gestation until delivery, the patient was on bed rest, Ibuprofen (2,400 g daily) was prescribed from 20 weeks until 1 week prior to delivery. At 35 weeks gestation, after a hospital stay and discharge of 24 h, the patient returned to the hospital in labor. Ultrasound confirmed fetal presentations to be vertex-vertex. Twin A (male, 2,400 g, Apgars 8/9) was delivered by spontaneous vaginal delivery. Twin B (male, 1,995 g, Apgars 4/9) was delivered by primary lower cesarean section after a cord prolapse with spontaneous rupture of membranes with the fetal vertex noted to be still high, umbilical arterial pH 7.17. The placenta with membranes was examined and found to be diamniotic dichorionic.

Conclusion

Improved pregnancy success rates following transfer of cryopreserved thawed embryos are expected with an increased number of embryos transferred. However, a decision on the number of embryos to transfer in both the retrieval and the frozen embryo cycle was necessary in this case, since there was concern that her unicornuate uterus would preclude a successful outcome if a multiple gestation occurred.

Unfortunately, after diagnosing twins by sonography, we could not find any reports in the English literature of multiple births in a unicornuate uterus to use as a precedent. The option of selective reduction was discussed with the patient [8, 9]. She was advised that we would have to consider her situation analogous to a quadruplet pregnancy. We related our personal statistics involving 32 patients with high-order multiple gestations offered reduction to a minimum of twins where all 14 patients choosing selective reduction had a successful outcome (13 of 14 babies delivered) versus only 18 of 25 not choosing selective reduction delivered at least 1 live baby [10]. Furthermore, there was a significant higher rate of prematurity in the latter group [10]. Despite these data, the patient declined selective reduction.

There appears to be an inherent bias in conclusions made from literature surveys on the unicornuate uterus. Many were reported in women with a history of recurrent abortion, yet the viable pregnancy rate in women with uterine anomalies were compared to the expected outcome in normal pregnancies and not in habitual aborters. There are many who believe that the unicornuate uterus is more common than is thought and might not even compromise a successful outcome. Certainly, most would agree that the septate or didelphys uterus represents a much more serious threat than the unicornuate uterus [11].

A successful outcome in this case report does not prove that subsequent cases of twins in a unicornuate uterus will produce at least 1 viable baby. Thus, the option of selective reduction should still be presented. The case described herein, however, at least presents a precedent for success that was not available to this patient when she had to make a decision.

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