An Incidental Finding of Unicornuate Uterus with Unilateral Ovarian Agenesis during Laparoscopy in Patient who Gave Birth to Eleven Children: A Case Report

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A B S T R A C T

Congenital uterine anomalies are often asymptomatic. They may present with infertility, recurrent miscarriage, preterm delivery, abnormal lie in pregnancy and other obstetric complications. We report the case of a 38-year old patient with unicornuate uterus without rudimentary horn and with unilateral left ovarian agenesis and unilateral left renal agenesis who gave birth to eleven children. Anomaly was incidentally diagnosed during laparoscopic sterilization.

***Key words**: unicornuate uterus, ovarian agenesis, renal agenesis, reproductive outcome, laparoscopic sterilization*

Introduction

Anatomic uterine defects have long been recognized as a cause of obstetric complications. The true incidence of congenital uterine anomalies is difficult to determine since many asymptomatic women with such anomalies are not diagnosed. Raga et al. found uterine anomalies in 4.0% of 3,181 women undergoing hysterosalpingography and laparoscopy/laparotomy[1]. Infertile patients had significantly higher incidence of müllerian anomalies (6.3%), in comparison with fertile (3.8%) and sterile (2.4%) women[1]. Septate (35%), bicornuate (25%) and arcuate (20%) uterus are the most common uterine anomalies. Arrested or defective development of only one müllerian duct results with a unicornuate uterus. A unicornuate uterus accounted for 9.6% of all uterine anomalies[2]. Incidence data in the literature reveal that unicornuate uterus occurs in 1:4020 women in the general population[3]. A unicornuate uterus without a rudimentary horn is present in 35% of patients with unicornuate uterus, a unicornuate uterus with noncavitary rudimentary horn in 33%, a unicornuate uterus with noncommunicating cavitary horn in 22%, and a unicornuate uterus with communicating cavitary horn in 10%[4]. The uterovaginal malformations are often associated with urinary tract anomalies and rarely with ovarian anomalies.

Case Report

A 38 years old female patient was admitted to the Division of gynecology and obstetrics, General hospital Bjelovar, Croatia, for laparoscopic sterilization. The patient was conscious, contactable, and cardiopulmonary compensated. She was mentally retarded and suffered from arterial hypertension, chronic renal insufficiency and duodenal ulcer. There was no family history of reproductive and urinary tract anomalies. She gave birth to eleven children during the period from 1986 to 2006, five at home and six in maternity hospital. During her pregnancies she did not have regular gynecological exams. Ten deliveries were term vaginal births. She did not have abortion. During her eighth pregnancy disturbances of renal functions and elevated blood pressure were diagnosed. The clinical and laboratory findings were con-

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firmed by ultrasound and radiologic examinations and final diagnosis of chronic renal insufficiency of unknown etiology with left renal agenesis was made. During her eleventh pregnancy she was undergoing hemodialysis. Her pregnancy ended in preterm delivery, and a few days after delivery this newborn child died. After her eighth pregnancy she could not avoid further pregnancies using available contraceptive methods and because of the complications during her eleventh pregnancy surgical sterilization was indicated. The laparoscopic sterilization was the technique of choice. At laparoscopy she was found to have unicornuate uterus with no evidence of a rudimentary horn and the absence of a left ovary, left fallopian tube and left round ligament. The right fallopian tube, right round ligament, and right ovary were all normal.

Discussion

We report the case of a patient with unicornuate uterus without rudimentary horn with unilateral ovarian agenesis and unilateral renal agenesis which was incidentally diagnosed during laparoscopic sterilization after eleven pregnancies and eleven childbirths.

Up to 40–50% of females with uterovaginal malformations have associated urinary tract anomalies[4], due to intertwined development of female reproductive system and urinary system. The close association between the mesonephric (wolffian) and paramesonephric (müllerian) ducts has important clinical relevance because developmental insult to either system is often associated with anomalies which involve the kidney, ureter and reproductive tract. Unilateral renal agenesis occurs when one or both ureteral buds fail to form or degenerate[5]. Because of the close embryologic association between the genital and urinary tracts, evaluation of the urinary tract in any patient with a genital anomaly is necessary. The association of anomalies of the müllerian ducts with concomitant ovarian anomalies is very rare because they develop from the other embryonic structures[4,6,7]. Unilateral ovarian absence may result from a congenital malformation (agenesis or aplasia) or from ovarian torsion with necrosis and reapsorption. The literature revealed only a few cases of unicornuate uterus with unilateral ovarian agenesis[6] and only two cases of unicornuate uterus with unilateral ovarian agenesis and with renal anomalies[7,8]. The pathophysiology of these rare combinations is not clear. The unicornuate uterus is associated with a poor reproductive outcome[1]. Fetal survival rates are poor. Reported live birth rate is from 29 to 49.9%. A review of literature revealed rates of 2.7–4.0% ectopic pregnancy, 24.3% first trimester abortion, 9.7% second trimester abortion, 20.1–44.0% preterm delivery, 10.5% intrauterine fetal demise[3,9]. Only 31.3% of all pregnancies end at term[1]. Obstetric complications such as breech presentation, fetal growth restrictions, dysfunctional labor and caesarean delivery are also more common[10]. Women presenting with a history of uterine anomaly should be considered high-risk obstetrical patients.

Conclusion

Most cases of anatomic uterine defects are diagnosed during evaluation for reproductive, obstetric or gynecological problems, but in the absence of these problems most anomalies go undiagnosed. In many patients, uterine congenital anomalies have been related with infertility, recurrent pregnancy loss, prematurity and other obstetric complications which increase perinatal morbidity and mortality. Our patient did not have conception, infertility or obstetric problems and there was no need for diagnostic procedure that would bring accurate diagnosis earlier. To our best knowledge this case is unique since the incidental diagnosis of unicornuate uterus with unilateral ovarian agenesis was made during laparoscopy sterilization after eleven pregnancies.

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SLUČAJAN NALAZ JEDNOROGE MATERNICE S JEDNOSTRANOM AGENEZIJOM JAJNIKA TIJEKOM LAPAROSKOPSKE STERILIZACIJE KOD PACIJENTICE KOJA JE RODILA JEDANAEST DJECE: PRIKAZ SLUČAJA

S A Ž E T A K

Urođene su anomalije maternice često asimptomatske. Mogu se očitovati neplodnošću, ponavljanim pobačajima, prijevremenim porodima, abnormalnim položajima fetusa i drugim porodničkim komplikacijama. Prikazujemo slučaj 38-godišnje pacijentice s jednorogom maternicom bez rudimentarnog roga, agenezijom lijevog jajnika i agenezijom lijevog bubrega koja je rodila jedaneaest djece. Anomalija je slučajno dijagnosticirana tijekom laparoskopske sterilizacije.