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BRIEF COMMUNICATION

Congenital Uterine Anomaly and Pelvic Organ Prolapse: A Rare Case of Pelvic Organ Prolapse in a Complete Bicornuate Uterus with Successful Pregnancy Outcomes Undiagnosed until the Time of Sacrocolpopexy

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Müllerian development anomalies (MDAs) are most commonly diagnosed in the reproductive period. A bicornuate uterus is the result of a fusion defect of the Müllerian ducts, causing an abnormal fundal outline. Most of the cases are diagnosed early in life and present with obstetrical complications, such as recurrent pregnancy loss, preterm birth, intrauterine growth restriction, placental abruption, and cervical incompetence. Pelvic organ prolapse (POP) in women with MDAs has been reported; however, all reported cases were when MDAs are diagnosed before or simultaneously with the development of POP in premenopausal young women aged < 35. A 52-year-old menopausal woman, who successfully delivered vaginally at term, was presented with protruding mass through vaginal introitus. On POP-Q examination, the cervix was elongated and descended to 1 cm out of the hymen during bearing down; however, the uterine bodies were confined in the pelvic cavity, which is commonly encountered among POP patients with large uterus due to uterine fibroids or adenomyosis. She also diagnosed for complete bicornuate uterus and underwent robotic sacrocolpopexy for advanced stage POP. It is presumed to have been caused by the bicornuate uterus that prevented the total uterine prolapse with the effect of extending both uterine horns bilaterally inside the pelvic cavity and trapping the uterus within the pelvis. Herein, we report a rare case of complete bicornuate uterus with multiple successful vaginal deliveries at term without obstetric complications, which remained undiagnosed until she was managed for the POP in her postmenopausal period.

Key Words: Pelvic organ prolapse, Pregnancy, Robotic surgical procedures, Uterine anomalies

INTRODUCTION

Congenital malformation of female genital tract is resulted from embryological maldevelopment of the Müllerian ducts (MDs) or paramesonephric ducts. Müllerian development anomalies (MDAs) arise whenever there is dysregulation in complex process of cellular differentiation, migration, fusion and canalization [1]. MDs differentiate to uterus, fallopian tubes, cervix, and upper one-third of the vagina. Failure in the development of MDs results in uterine agenesis, and incomplete development leads to uterine hypoplasia or a unicornuate uterus. Incomplete fusion of the MDs results in uterus didelphys, bicornuate uterus, and arcuate uterus [1]. MDAs are rare with a prevalence of 4%–7% [1]. Bicornuate uterus represents one-fourth of MDAs [2]. MDAs have been found to be associated with pregnancy loss, preterm birth (PTB), malpresentation, and cesarean delivery [2]. In women with bicornuate uterus, conception does not appear to be affected; however, there is an increased risk of obstetrical complications such as recurrent pregnancy loss, PTB, intrauterine

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growth restriction (IUGR), placental abruption, and cervical incompetence [2].

In terms of pelvic organ prolapse (POP), the main cause of POP is weakening of the supportive ligaments. The multiparity and increased age certainly are risk factors in the development of genital prolapse [3]. Classification of POP consists of anterior compartment, apical compartment, posterior compartment prolapse. Anterior compartment prolapse is the most common form and the old aged women usually manifested as multi-compartment POP, whereas young aged women have mostly one compartment prolapse usually an apical compartment POP [3].

Diagnosis of MDAs associated with POP in the old age is rare. Among the publications reported MDAs associated with POP to date, 35-year-old patient was the most oldest woman [2,4]. MDAs are usually diagnosed during evaluation due to symptoms such as abdominal pain and amenorrhea, and without symptoms, problems with obstetric outcome may have occurred. Here we report a rare case of complete bicornuate uterus which was not diagnosed even in pregnancy and childbirth. She carried her pregnancies to term and delivered by vaginal normal deliveries without any obstetric problems or complications. She first became aware of bicornuate uterus when diagnosed as POP with apical prolapse. An uneventful robotic sacrocolpopexy was done and POP has been completely resolved at her follow-up at postoperative 6 weeks. This is a first case report of POP associated with bicornuate uterus in a multiparous, postmenopausal woman without any obstetric complications.

CASE REPORT

This study was conducted according to the guidelines of the Declaration of Helsinki (2013) and was approved by the Asan Medical Center Institutional Review Board (approval No. 2021-0292).

A 52-year-old menopausal woman (gravida 5, para 2) presented with intermittent dysuria, stress urinary incontinence and bothersome protruding mass through the vaginal introitus for several years which was aggravated for 1 year. She got pregnant naturally and had a history of two uncomplicated spontaneous vaginal deliveries without dystocia, malpresentation of baby, and IUGR at term. Her obstetric history was 2-0-3 (1 spontaneous abortion at early pregnancy, 2 artificial abortion)-2. When she looked back, she only remember one obstetrician's opinion that her uterus, which seems something different from normal uterus. However, successful normal pregnancies and normal deliveries in women with complete bicornuate uterus are extremely rare. At the first visit, pelvic examination revealed a one vagina, one cervix and POP-Q stage III (+3, +5, +1, 0, 0, –3) (6, 4, 8) based on the Pelvic Organ Prolapse Quantification System (POP-Q). The anterior vaginal wall was fully prolapsed and the most distal portion of the cervix descent 1 cm below the plane of the hymen. There were no signs of rectocele and enterocele on translabial sonography. On transvaginal sonography, we found two horns of uterus, which the patient recognized it for the first time. Urodynamic study revealed the urodynamic stress incontinence. The patient underwent robotic

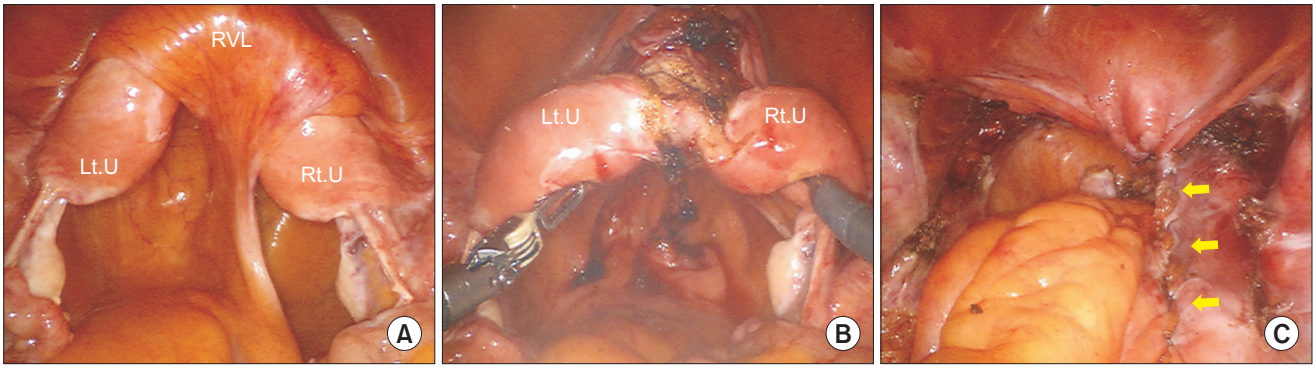

Fig. 1. Laparoscopic view. (A) The rectovesical ligament was crossing the junction between two horns of uterus. (B) After dissection of rectovesical ligament from bladder and rectum, indentation at the level of uterine midline fully dividing uterine corpus was seen. (C) Robotic subtotal hysterectomy, bilateral salpingo-oophorectomy, and sacrocolpopexy with retroperitoneal tunneling technique (arrows) were performed. Lt.U: left hemiuterus, Rt.U: right hemiuterus, RVL: rectovesical ligament.

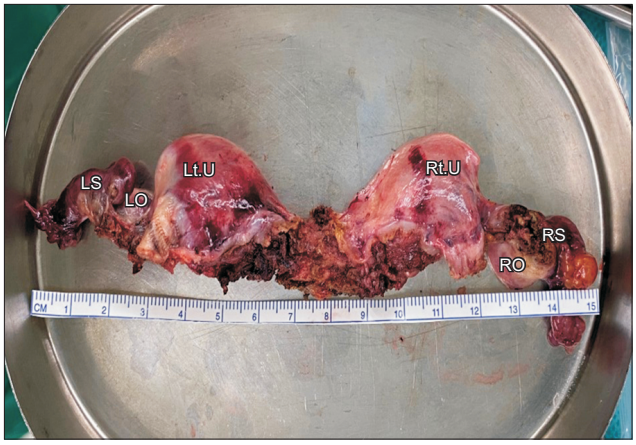

Fig. 2. Postoperative specimen showing bilateral uterine horns with normal ovaries and fallopian tubes. LS: left salpinx, LO: left ovary, Lt.U: left hemiuterus, Rt.U: right hemiuterus, RO: right ovary, RS: right salpinx.

supracervical hysterectomy, bilateral salpingo-oophorectomy, sacrocolpopexy and transobturator vaginal tape insertion. Robotic sacrocolpopexy was performed as described in our previous reports [5-7] using the da Vinci Xi system (Intuitive Surgical, Sunnyvale, CA, USA). Intraoperative findings included a rectovesical ligament crossing the junction between two horns of uterus and normal fallopian tubes and ovaries (Fig. 1A). After dissection of rectovesical ligament from bladder and rectum, indentation at the level of uterine midline fully dividing uterine corpus was seen (Fig. 1B). In the postoperative specimen, we could definitely identify two uterine horns with normal ovaries and fallopian tubes (Fig. 2). The postoperative course was uneventful. At the 6 weeks of follow-up visits, the patient had complete resolution of prolapse with POP-Q stage 0 and reported that dysuria and stress urinary incontinence had disappeared.

DISCUSSION

Congenital genital tract malformations are not common, and bicornuate uterus is one of the common type of MDAs [1]. The reliable classifications have been proposed because MDAs are associated with health and reproductive problems depending on the type and degree of anomalies. The American Fertility Society (AFS) categorized female genital tract anomalies in 7 classes [8]. Bicornuate uterus is defined as double uterus with a single cervix, and divided into two subclasses, partial and complete, according to the degree of the uterine body deformity [8]. The European Society of Human Reproduction and Embryology (ESHRE) developed an updated classification system [1]. Class U3b or complete bicorporeal uterus is defined the uterus with an external fundal indentation completely dividing the uterine corpus up to the level of the cervix, as in this case. The obstructive MDAs are mostly undiagnosed until the patient presents with hematometra or hematocolpos; however, a septate uterus or bicornuate uterus are mostly asymptomatic and diagnosed in patients with infertility or pregnancy. In addition, the diagnosis can be missed at the time of antenatal care, if nongravid uterus is positioned posterior to gravid uterus, as in this case [2].

It has been established that MDAs are related to adverse pregnancy outcomes. The presence of uterine anomalies increased risk in recurrent pregnancy loss, PTB, preterm premature rupture of membranes, breech presentation, cesarean delivery, placenta previa, placental abruption, and IUGR [9]. It is hypothesized that abnormal uterine blood flow and decreased muscle mass would be reasons for PTB or IUGR [9]. Grimbizis et al. [10] reported a spontaneous abortion rate of bicornuate uterus as 36% and a PTB rate of 23%. In addition, Heinonen et al. [11] identified that the deeper the bifurcation the poorer the obstetric performance. The incidence of PTB was 29% in partial and 66% in complete bicornuate uterus. Breech presentation of baby was 0% in partial and 50% in complete bicornuate uterus, and cesarean delivery rate was 20% and 36%. However, there is a disputing result that patients with the partial type of bicornuate uterus showed favorable outcomes than complete type for achieving pregnancy and childbirth following metroplasty procedure [12]. Our patient had a first trimester spontaneous abortion, but there were no adverse pregnancy outcomes such as uterine rupture, PTB, and cesarean delivery without correction of the bicornuate uterus by metroplasty surgery. Cesarean section is commonly recommended to women with bicornuate uterus for the risk of uterine rupture before or during the labor. However, in this case, successful vaginal deliveries were accomplished at term.

It is well-known that multiparity and advanced age are major risk factors for POP [13]. Despite these associations, the primary site of older age group was distributed evenly between different sites. The multicompartment prolapse and a higher grade of prolapse in older women may represent that the first

site of prolapse is at the apical support, and the other connective tissues fail to support over time [3]. Some authors explain that the upper portion of the vagina is derived from Müllerian structures, so that loss of apical support in young patients with MDAs was related to aberrant uterine anatomy [14]. However, in our patient, the primary site of prolapse was anterior compartment with elongated cervix. The elongated cervix with uterine fundus remained in the pelvis was noted in this case. It is probable that bicornuate uterus made an effect of support by uterine horns extending to both sides of pelvis and trapped in the pelvis.

In this case, the uterine anomaly was diagnosed late in her age during management of POP. When a search of PubMed was performed for all studies published from January 1986 to January 2021 using terms 'Müllerian development anomalies' and 'pelvic organ prolapse', there were 19 reports of MDAs diagnosed before age of 35 associated with POP. Fourteen of the 19 reports were cases in which neovagina was prolapsed after being diagnosed and treated as MDAs. The neovaginal prolapse is considered that the surgical creation of vagina allows for normal menstruation and sexual function but does not provide endopelvic facial support of neovagina [15]. POP and MDAs were diagnosed simultaneously in five of the 19 reports. Longitudinal vaginal septum associated with POP in a 35-year-old multiparous woman was reported [4]. The patient was not aware of the vaginal anomaly. Her uneventful three normal vaginal deliveries were conducted with untrained birth attendant. There is the case of a 17-year-old primigravid patient with bicornuate uterus and cervical prolapse at 38 weeks of gestation. She was delivered with cesarean section due to obstruction of the lower uterine segment of the gravid uterus [2].

To our knowledge, this is the first report of POP occurred in a bicornuate uterus with successful pregnancy outcomes without surgical correction of MDAs. In this case, women with a complete bicornuate uterus can conceive spontaneously and successfully give a birth vaginally without midterm adverse pregnancy outcomes. Anatomical variants of MDAs such as complete type of bicornuate uterus may contribute to the clinical manifestations of the POP.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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