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An Uncommon Mullerian Anomaly Without Classification: Septate Uterus Cervical Duplication and a Longitudinal Vaginal Septum

Tahereh Behrouzi-lak*, Somayeh Ghasemzadeh

Reproductive Health Research Center, Gynecology Department, Urmia University of Medical Sciences, Urmia, Iran

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Corresponding Information:
Tahereh Behrouzi-lak,
Reproductive Health Research Center, Gynecology Department, Urmia University of Medical Sciences, Urmia, Iran
Email: t.behrooz2@yahoo.com

ABSTRACT

The most common form of structural uterine anomaly is the septate uterus, which has the highest rate of reproductive failure. An uncommon type of development is a septate uterus with cervical duplication and a complete longitudinal vaginal septum. We report a 26-year-old woman with infertility complaints. We performed diagnostic procedures and noticed that the patient had a septate uterus and cervical duplication with a longitudinal vaginal septum. It is an uncommon Mullerian anomaly that does not fit into the current assortment system of Mullerian anomalies by the American Fertility Society (AFS).

Keywords: Mullerian anomaly, Septate uterus, Vaginal septum

Introduction

Congenital uterine malformations result from disruptions in the development, formation, Mullerian fusion, or para-mesonephric ducts during fetal life **(1)**. The incidence of uterus congenital malformations in the general population is 0.001–10% **(2)**. The prevalent form of the structural uterine anomaly was the septate uterus, with the highest reproductive failure rate. A rare type of development is a septate uterus with cervical duplication and a complete longitudinal vaginal septum. This anomaly was first introduced by McBean and Brumsted **(3)**. The existence of this anomaly challenges the classical theory of the Mullerian developmental **(4)**. The traditional embryologic hypothesis is that Mullerian development proceeds in a unidirectional (from caudal to cranial fashion), and as a result, the septum is later absorbed. **(5)**. We report the case of a woman with a septate uterus and cervical duplication and a longitudinal vaginal septum that does not fit into the current assortment system of Mullerian anomalies by the American Fertility Society (AFS).

Case Report

A 26-year-old woman, Gravida 2 Aborted 2, married for 4 years presented with complaints of secondary infertility. In the history taken from the patient, her menarche happened at age 14. She has regular menstrual cycles with intervals of 28 to 31 days and a duration of 4 to 6 days. The patient had no history of dysmenorrhea and dyspareunia. There was no history of pelvic infection disease, abnormal uterine bleeding, and no complaint of excess waste hair growth and secretion from the breast. She has been pregnant spontaneously two times and aborted for less than ten weeks. She had no history of specific medical diseases or surgery. Gynecological examination revealed a longitudinal vaginal septum that extended to the cervix with two separate cervical os that the left cervical os was smaller than the right. Transvaginal ultrasound showed a normal uterine characteristic and the adnexes were normal (Figure 1), then hysterosalpingography was performed. In hysterosalpingography, they were unable to inject contrast media from the left cervix and injection from the right cervix showed that right tube was open (Figure 2). The patient underwent a laparoscopy and hysteroscopy. On laparoscopy, a single fundus of uteri with normal adnexa was observed. endometriotic lesion was evident in the

middle part of the right fallopian tube and the left ovarian surface (Figure 3). On hysteroscopy with findings of a complete septate uterus with double cervix and a vertical vaginal septum that the left side was smaller than the right (Figure 4). The depth of the uterine cavity was 8 centimeters. During the laparoscopy, Methylene blue was injected through both cervices respectively. By injecting methylene blue from the right cervix, there was a fall in the right tube, but when injected into the left cervix, there was no drop in the left tube.

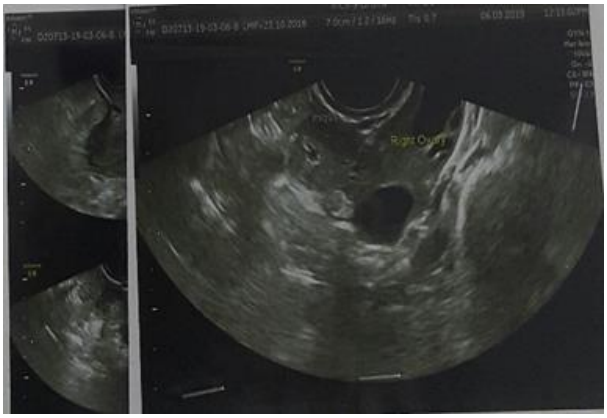

Figure 1. Transvaginal ultrasound

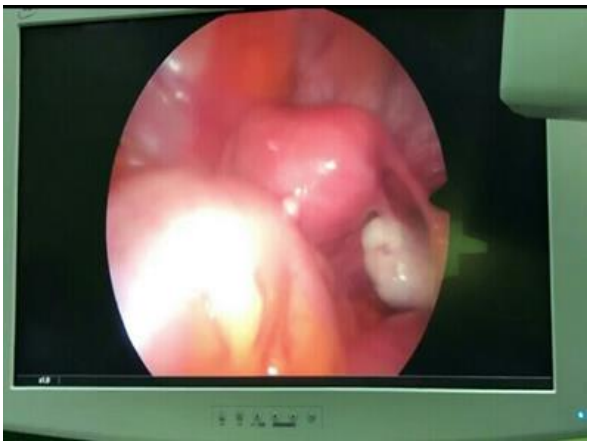

Figure 2. Laparoscopy

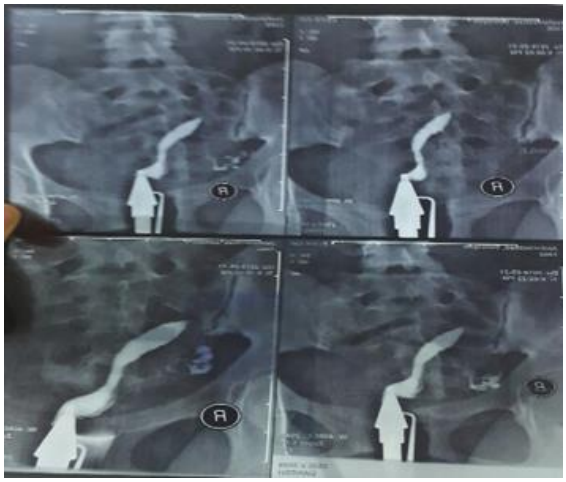

Figure 3. Hysterosalpingography

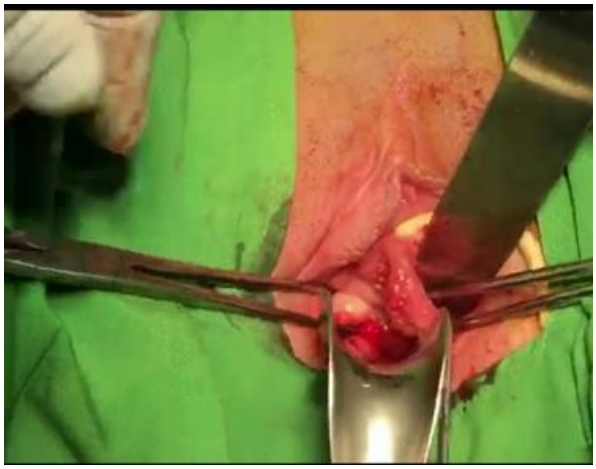

Figure 4. Vertical vaginal septum

Discussion

Mullerian abnormalities consist of morphological, embryological basis, clinical, and functional criteria. **(6)**. McBean and Brumsted **(3)** reported the unique Mullerian anomaly consisting of a septate uterus with cervical duplication and longitudinal vaginal septum for the first time. This anomaly is distinct from the classical embryological definition of Mullerian anomalies. The currently accepted theory cannot explain the case introduced here. This case fits into an alternate approach, proposed by Musses in 1967, where the uterus and cervix and upper vagina develop by a three-stage process, in which medial aspects of the Mullerian ducts begin to fuse in the middle and proceed both caudad and cephalad directions simultaneously **(7)**. A rare number of similar anomalies have been reported with different symptoms. Some cases have never been diagnosed or have been misdiagnosed **(8)**. As it is known, in an abnormal uterus, abortion and abnormal presentations are more common than normal uterus, and the septate uterus is not an infertility factor in itself. In one of the reported cases, pregnancy occurred with Intrauterine Insemination (IUI) in two cavities of the uterus **(9)**. The approach to managing this anomaly is controversial. It seems that hysteroscopic resection of the septum is the gold standard to improve obstetric outcomes, as this method has been used in reported cases **(10-11)**.

Conclusion

The case reported here is a rare type of Mullerian anomaly that cannot be explained in the usual classification. These cases need to be followed, and an evaluation of family history is required to determine its effect on fertility and develop optimal treatment options.

Acknowledgments

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Conflict of Interest

The authors declared no conflict of interest.

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