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Case Report

Septate uterus with cervical duplication and a longitudinal vaginal septum: an unclassified mullerian anomaly

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ABSTRACT

Uterine anomalies are the result of disturbances in the development, formation or fusion of the mullerian ducts during foetal life with an incidence of 0.001%-10% in general population. These patients most often present with amenorrhoea, dysmenorrhoea, dyspareunia and infertility. Women with these anomalies are more prone to poor foetal outcomes such as first trimester losses or preterm labour. We report a case of a 35 years old female with septate uterus with cervical duplication and a longitudinal vaginal septum. Diagnosis was obtained by physical examination, ultrasound imaging, hysterosalphingography and magnetic resonance imaging and hysterolaproscopy. The clinical presentation of such a case & surgical options available were studied, and the excision of the vaginal septum was done. This case of congenital uterine anomaly does not fit into the current classification system of mullerian anomalies by American fertility society, which is based on the traditional caudal to cranial, unidirectional developmental pattern. This strongly indicates the possibility of an alternative mechanism of mullerian development.

Keywords: Septate uterus, Cervical duplication, Longitudinal vaginal septum

INTRODUCTION

Congenital uterine malformations are the result of disturbances in the development, formation or fusion of the mullerian or para-mesonephric ducts during foetal life. The incidence of congenital uterine anomalies in the general population is 0.001%-10%.[1] Patients with congenital uterine malformations might present with amenorrhoea, dysmenorrhoea, dyspareunia and infertility. Women with these anomalies are more prone to poor fetal outcomes such as first trimester losses or preterm labour.[2]

The traditional hypothesis is that mullerian development proceeds in a unidirectional, caudal to cranial fashion and the resulting septum is later absorbed. Throughout the recent literature, cases have been reported that do not seem to follow this development pattern. We report a case of unique congenital uterine anomaly that does not fit into the current classification system of mullerian anomalies by American fertility society, thus further strengthening the possibility of an alternative mechanism of mullerian development.[3,4]

CASE REPORT

A 35 year old nulliparous woman presented with menorrhagia, dyspareunia for 5 years and secondary infertility for 10 years. She has been married for 11 years and had one spontaneous complete abortion at 3 months of gestation, 10 years back. The patient was obese with BMI of 35, had hirsuitism. No endocrinological abnormality was found on investigation. Gynaecological examination revealed a vertical vaginal septum with two normal-appearing cervices (Figure 1). Transvaginal ultrasound examination demonstrated one uterine fundus with two uterine cavities. A hysterosalpingogram confirmed two cervical openings and two separate uterine

cavities. A subsequent magnetic resonance image failed to reveal a fundal cleft, excluding a uterus didelphys (Figure 2). Intravenous pyelography was normal. The patient underwent a laparoscopy and hysteroscopy, with findings of a complete septate uterus with double cervix and a vertical vaginal septum. Chromotubation done with both the uteri and revealed bilateral tubal spill. The vaginal septum was resected to relieve dyspareunia and to improve access to the left cervix, which was limited by a narrower hemivagina (Figure 3). The symptomatic complaint secondary to the vaginal septum got resolved. The case is being followed up for further management in case she conceives or otherwise as the case may be.

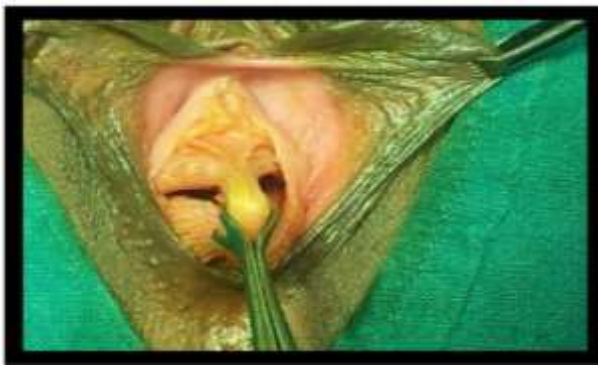

Figure 1: Vertical vaginal septum with two normal appearing cervices.

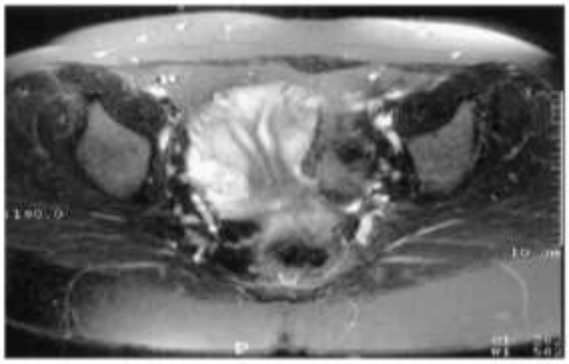

Figure 2: MRI image.

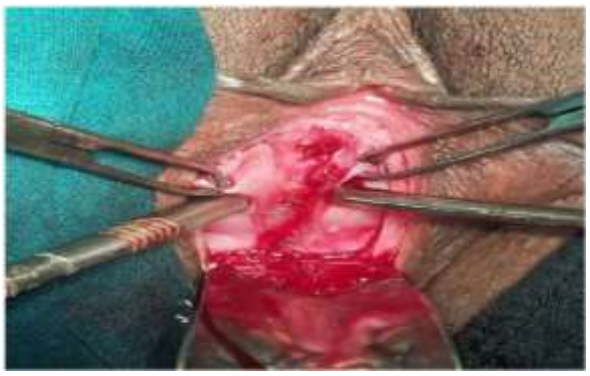

Figure 3: Two cervices seen after resection of vaginal septum.

DISCUSSION

Rare anomalies pose a challenge to the practitioner in making the correct diagnosis and deciding upon the therapeutic options. Lin PC et al reviewed that MRI correctly differentiated the type of mullerian anomaly in 96% of patients. In addition, MRI allows for evaluation of the urinary tract.[5] Acien P studied that urinary tract abnormalities were present in 30% of women with mullerian anomalies.[6] Vaginal septum is associated with uterine malformations in 81% of cases.

The choice of therapeutic modality presents a challenge in this anomaly. Previously, resection of the uterine septum was advocated.[7] However, in the absence of history of recurrent pregnancy loss, it was decided that the increased risk of uterine perforation, scarring or cervical incompetence did not warrant the additional procedure.[8] Moreover septate uterus is not an infertility factor in itself. In addition, there are reports of similar cases documenting full term delivery after resection of the vaginal septum only.[3,10]

In view of increased number of similar unique anomalies being reported, in 1967 Muller et al proposed that fusion begins at the level of the isthmus and proceeds simultaneously in both the cranial and caudal directions. Development of the uterine corpus is followed by resorption of the septum, which begins at the isthmus and proceeds in both directions. This theory could account for a normal fundus in a patient with an otherwise duplicate uterus and upper vagina. It would also account for the findings described in the cases cited above. In light of these recent cases, the addition of a classification category allowing for alternative developmental models might now be appropriate.[9]

CONCLUSIONS

The classical embryological model forms the basis of classification system of mullerian anomalies by Buttram and Gibbons, which is currently used by American fertility society.[11] However, in view of reports of similar unclassified anomalies an alternate development mechanism is suggested.[3,4,7] Thus, rare anomaly is not explained by the traditional embryological teaching and not described in the currently used classification system for mullerian anomalies. These unique cases need to be followed longitudinally so that its effect on fertility can be determined and optimal treatment options can be developed.

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