Case Report

Bicervical Normal Uterus with Normal Vagina

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Abstract

This is a report of the form of uterine anomaly involving a dual cervical canal in a side-by-side disposition with normal uterine cavity and normal vagina. It portrays a form of congenital uterine anomaly not explicable by the existing classical theory of mullerian anomalies. However, there has been a proposed reclassification of mullerian anomalies, which includes this type of anomaly under Type IIIc. The patient was a 31-year-old woman being managed for "secondary infertility." To report a case of uterine anomaly that is not explicable by the existing classical theory of mullerian anomalies.To the best of our knowledge, only few cases of bicervical normal uterus with normal vagina exist in the literature; one of the cases had an anterior-posterior disposition. This form of uterine abnormality is not explicable by the existing classical theory of mullerian anomalies and suggests that a complex interplay of events beyond the classical postulate gives rise to the female genital tract.

Keywords: Anomaly, Bicervical, Uterus

Introduction

The embryology of the female genital tract classically involves a complex sequence of events leading to differentiation, migration, caudo-cranial fusion, and canalization of the mullerian ducts and urogenital sinus. Any dysregulation or interruption of this process can lead to a wide range of mullerian duct anomalies.[1,2] The occurrence of some mullerian anomalies not explicable by the classical theory led to alternative postulations which favor a segmental and bidirectional fusion of the mullerian ducts,[3,4] and divergence of the fused ducts followed by re-fusion.[5,6]

The case being presented and the articles by Morales-Roselló and Peralta Llorens[7] and Acién *et al*.[8] support these alternative hypotheses.

Case Report

A case of 31-year-old woman with secondary infertility, referred to our department for hysterosalpingography. She attained menarche at 13 and had regular 28-day menstrual cycle of 4-day menstrual flow. There was a history of term gestation that resulted in stillbirth following prolonged labor.

A hysterosalpingography was performed, and it was observed that contrast was tracking out through a second cervical os as the uterine cavity is being filled with a contrast. The images taken showed a second cervical canal that was adjacent and lateral, but similar in caliber to the cannulated cervix [Figure 1]. The double cervical canals were also clearly demonstrated on the transvaginal sonography [Figure 2]. They joined at a common internal os before opening into a normal uterine cavity. Neither a septum nor another abnormality was detected in the vagina.

Discussion

In females, mesoderm lateral to the mesonephric ducts give rise to the paramesonephric (Mullerian) ducts in the 7th week of the embryonic life. Initially, these ducts grow caudally, on the lateral side of the urogenital ridges, but cross medial to them by the 8 weeks. The distal portions of both paramesonephric ducts fuse in the midline to give rise to the upper two-third of the vagina, the cervix, and the uterus. The unfused proximal cranial portions remain as the oviducts (fallopian tubes). The entire process involves a complex, but coordinated cascade of events, which disruption at any point would result in developmental anomalies of the female genital tract.

Mullerian anomalies are not uncommon, especially in women presenting with reproductive complications. Incidences as high

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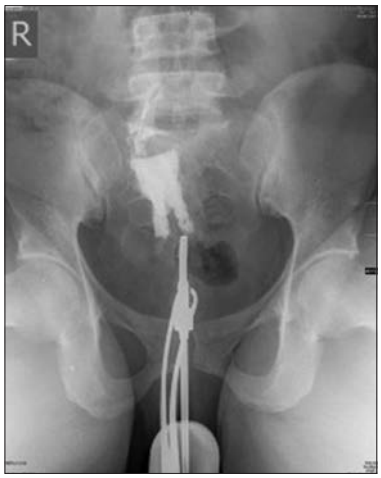

Figure 1: Hysterosalpingograph showing double cervix in the 31-year-old female being investigated for secondary infertility

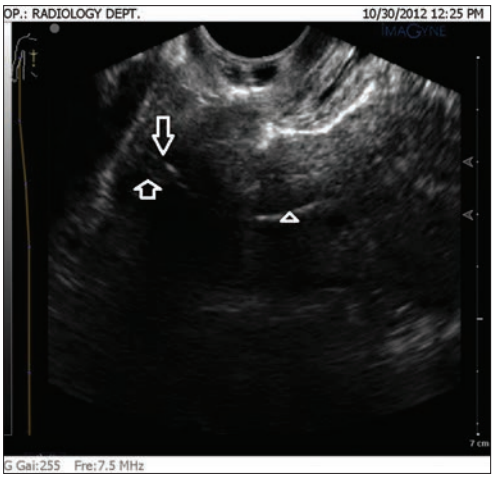

Figure 2: Transvaginal ultrasound image of the uterus in the same patient. Arrows show endocervical stripes. Triangle shows endometrial stripe

as 5–10% have been documented in women having recurrent abortions and third-trimester pregnancy losses.[9,10] In the general female population, it is, however, less with a mean incidence of 4.3% reported by some authors.[11]

The most common forms of mullerian abnormalities are septate, arcuate, didephys, unicornuate, and hypoplastic uteri; with the precise incidence of each varying with locality.[1] Almost all of these anomalies are explicable by the classical theory of caudo-cranial fusion of the distal mullerian ducts. This theory, however, fails to explain the occurrence of double cervical canals with normal uterine cavity and normal vagina. This type of anomaly can be explained if considered in the light of Acien's hypothesis[5,6] that is, as the paramesonephric ducts are completing their caudal to cranial fusion into a single tube, the already fused most caudal portion diverges. The proximal and distal limits of this point of divergence correspond to the internal and external os, respectively. Hence, the point of divergence gives rise to the cervix. The part of the fused paramesonephric duct distal to the divergence fuses with the urogenital sinus to form the vagina, while the cranial fused portion gives rise to the uterus. The unfused most cranial parts of the mullerian ducts remain as the oviducts.

The defect in the case we report might have resulted from a defective re-fusion after the divergence. The clinical implication of the anomaly is that there may be ineffective dilatation and shortening of the cervix during labor with resultant prolonged labor. This may be responsible for our patient's prolonged labor, resulting in stillbirth.

The case clearly points out that the classical theory of female genital development is inadequate in explaining certain mullerian anomalies and gives further credence to Acien's postulate.

There has been other reclassification of Mullerian anomalies recently as highlighted in the study by El Saman *et al*.[12] The proposed reclassification placed our case under Class IIIc.

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