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Case Report

A rare case of double cervix with normal uterus and vagina: Müllerian anomaly in North India

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ABSTRACT

Bifid cervix is a rare Müllerian duct anomaly. We present a case of a 38-year-old woman with secondary infertility, found to have a cervical duplication. To the best of our knowledge, only a few cases of bicervical normal uterus with normal vagina exist in the literature. This form of uterine abnormality is not explicable by the existing classical theory of Müllerian anomalies and suggests that a complex interplay of events beyond the classical postulate gives rise to the female genital tract.

Keywords: Bifid cervix, Congenital anomalies, Double cervix, Müllerian anomalies, Müllerian duct malformations, Rare case

INTRODUCTION

Müllerian duct anomalies (MDAs) occur approximately in 5–7% of the general female population. Müllerian duct anomalies (MDAs) are frequently encountered and can result in issues related to menstrual flow, pregnancy, and fertility.¹ Two cervical canals, extending from the internal to the external os, denote a double cervix. The reproductive and surgical consequences of Müllerian anomalies are influenced by accurate diagnosis and classification.² The embryological development of the female genital tract traditionally encompasses a series of intricate processes, including differentiation, migration, caudo-cranial fusion, and canalization of the Müllerian ducts and urogenital sinus. Any unsettling or alteration in this sequence can result in various Müllerian duct anomalies.³ Surgical treatment for each type of Müllerian duct anomaly is categorized based on the classification provided by the American Society of Reproductive Medicine (2021), which encompasses conditions such as cervical agenesis, unicornuate uterus, uterus didelphys, bicornuate uterus, septate uterus, longitudinal vaginal septum, transverse

vaginal septum, and complex anomalies.⁴ We present a case of cervical duplication occurring alongside a normal uterus and vagina, but with an anteroposterior disposition of the cervix, which supports the theory that isolated segment defects may occur. This case challenges the embryological theories that have underpinned the American Society for Reproductive Medicine classification for many years.

CASE REPORT

A 38-year-old woman, P2L2, reported in outpatient department (OPD) with a chief complaint of abnormal uterine bleeding for the last 3 years. The patient has been on medical treatment since then, but her complaint has not been resolved. Menarche was at the age of 15 years, with regular menstrual cycles. She was sexually active. She denied any dyspareunia and the absence of any miscarriages. She had a previous 2 caesarean sections. Her past medical history is uneventful. A gynaecological examination per vagina revealed the uterus to be anteverted, normal size, the cervix firm, bilateral fornices

free and non-tender, with normal external genitalia and vagina. On ultrasound, the uterus was bulky. The patient was planned for an abdominal hysterectomy. Uterus and cervix removed. Two cervical orifices were clearly visualized, which was an incidental finding. In both cervices, an evident external os surrounded with a reddish endocervical epithelium was present. The consent has been taken by the patient for writing the report and uploading the pictures.

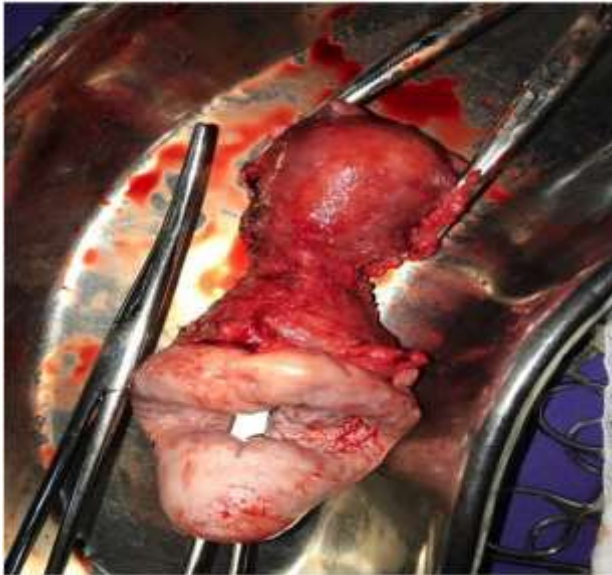


Figure 1: Intraoperative finding of the patient showing a bifid cervix.

DISCUSSION

Congenital anomalies of the female reproductive system are delineated as variations from normal anatomy arising from improper embryonic development of the Müllerian or paramesonephric ducts.⁵ In our case, we present an incidental finding of double cervix post hysterectomy in contrast to the case report by Antunes et al regarding a double cervix with a normal uterus and vagina, whose case had secondary infertility and the two cervical orifices were clearly visualized on gynecological examination. Also, the imaging in our case showed a bulky uterus with a normal cervix and vagina: however, the ultrasonography of the case by Antunes et al showed a normal uterus with two endocervical canals.

The most widely utilized classification system to date, introduced by the American Society for Reproductive Medicine in 1988, provides a thorough categorization of uterine anomalies but does not address abnormalities of the cervix or vagina. More recently, the European Society of Human Reproduction and Embryology has established a consensus that permits independent classification of cervical anomalies.⁶ The cause of most uterine anomalies in the female genital tract remains unclear. Conducting familial studies is challenging since some women with

these anomalies may be asymptomatic and still have a normal reproductive history.⁷

Two primary theories, both introduced in the 1960s, serve as the basis for the ASRM classification system. The first theory, proposed by Crosby and Hill, posits that the development of the uterus occurs through the fusion of the Müllerian ducts between the 11th and 13th weeks of embryonic development, starting from the caudal end and moving cranially. This process is escorted by the reabsorption of any septa that form, which can occur starting from any point of fusion and can proceed in one or both directions. A limitation of this unidirectional theory is that it fails to explain lower segment defects that coexist with normally formed upper segments, such as in cases of vaginal or cervical duplication alongside a normal uterus.⁸

The second theory proposes that Müllerian duct fusion begins in the middle portion at the uterine isthmus and occurs simultaneously in both cranial and caudal directions. It argues that septal reabsorption also follows a bidirectional pattern, resulting in complete uterine formation that is independent of cervical and vaginal development. Although this theory appears to address some defects that the first theory does not account for, it still does not clarify the mechanism behind isolated defects, as displayed in our case.⁸ The presented case aligns with Acien's theory, as the defect originates at the internal cervical os and extends to the external cervical os. There were no abnormalities detected in the uterus, and the lower portion of the fusion process was completed properly, resulting in a normal vagina.⁹

CONCLUSION

The wide variety of congenital Müllerian malformations underscores the complex nature of reproductive embryology and the potential disruptions that can arise during development, prompting a reevaluation of established concepts regarding embryological processes. As more unique and unclassified cases of Müllerian duct anomalies are identified, there is a growing push for revised classification systems that aim to improve the precise identification of defects and enhance patient care. This case report exemplifies one of the numerous abnormalities of the female reproductive tract that older embryological theories fail to address, thereby supporting modern theories and illustrating the intricate interactions involved in the development of the female genital tract. Our understanding of the embryology of the female genital tract remains incomplete, as Müllerian development is more complex than originally believed. Acien's segmentary theory provides the most convincing explanation for segment malformations, as demonstrated in this case, and prompts a reassessment of our long-standing views on embryological development.

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