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Case Report

An unusual complex mullerian anomaly: case report

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ABSTRACT

Congenital anomalies are a result of defect in the normal process of embryonic formation of organs. When more than 1 complete or incomplete defect coexists, they lead to formation of a complex anomaly. Here we report a case of an unusual and unclassified complex uterine anomaly and its management. To the best of our knowledge such a case has not been reported till date in the literature. The authors also reinstate the need for classifying these unusual anomalies.

Keywords: Congenital, Classification, Mullerian duct, Uterine anomalies

INTRODUCTION

Congenital uterine anomalies result from abnormal formation, fusion or resorption of the Mullerian ducts during fetal life. Complex anomalies are caused due to defect in more than one developmental lines. These are brought into light with complaints of infertility, miscarriage, premature birth, abnormal fetal presentation, cyclic pain, or as incidental diagnosis during pelvic or ultrasonography examination. The prevalence rates of uterine anomalies have varied between 0.06% and 38%.[1-8] This wide variation is due to the assessment of different patient populations and the use of different diagnostic techniques with variable accuracy rates as well as reliance on non-standardized classification systems. Combined (hysteroscopy/laparoscopy) is considered as the gold standard in diagnosing and treating congenital uterine anomalies.[7,8] To the best of our knowledge such a case has not been reported till date in the literature, adding to the spectra of complex anomalies.

CASE REPORT

We present a case of 26 year old woman married for 4 years. She had a history of medical termination of pregnancy 1.5 years back at 6 weeks gestation. She was also a known case of hypothyroidism. She had been trying to conceive for last 1 year. Her fertility work up done was perfectly normal except unicornuate uterus with unilateral spill in hysterosalphingography (Figure 1). Her ultrasound was normal. She was advised combined laparoscopy with hysteroscopy approach. Her hysteroscopy showed a unicornuate cavity with absence of right ostia (Figure 2). But to our surprise, her laparoscopy showed a normal shape and size of the uterus with unilateral deficient cavity. Half of the uterus was solid and half had the cavity (Figure 3). Bilateral ovaries were normal. The right sided fallopian tube was non-canalized (Figure 4). We did a hysteroscopic metroplasty for her to increase the cavity and make it more roomy. This would facilitate her for further planning her family. This case according to the new ESHRE classification comes in class VI of unclassified anomalies.

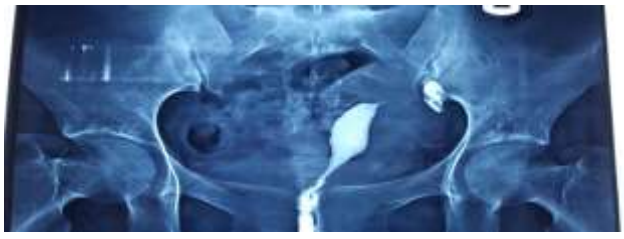

Figure 1: HSG showing unicornuate uterus with unilateral spill.

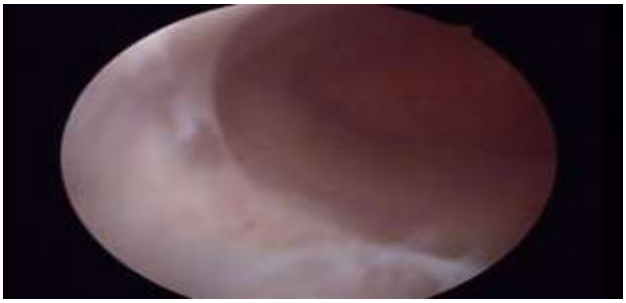

Figure 2: Hysteroscopy showing unicornuate cavity with 1 ostia.

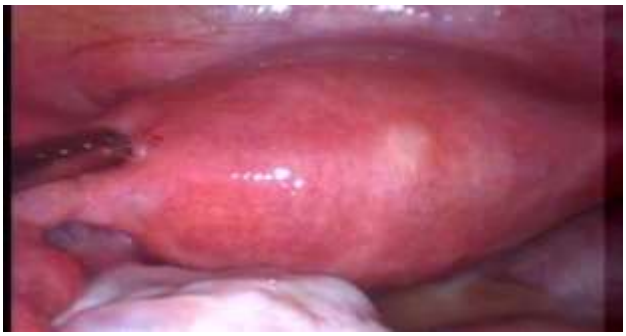

Figure 3: Normal size and shape uterus with hemi cavity.

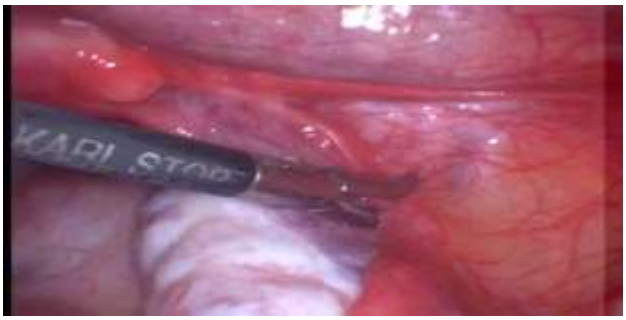

Figure 4: Righted rudimentary tube with normal ovary.

DISCUSSION

Complex uterine anomalies can occur due to complete or incomplete combination of two mullerian anomalies. These anomalies have been associated with an increased rate of infertility, miscarriage, preterm delivery and other adverse fetal outcomes.

Till now there is no classification which can describe all possible mullerian anomalies. Many classification systems are given to classify Mullerian duct anomalies with variable level of acceptance and For long the American Fertility Society (AFS) currently known as American Society of Reproductive Medicine system (ASRM) classification was the most accepted worldwide, being easy to interpret.[9-15] Sadly, it also had some fallacies, mainly difficult classification of mixed uterine anomalies, absence of diagnostic parameters for anomalies, complete dependence on subjective impressions of clinician performing the diagnostic test, being noncomprehensive to some rare anomalies and some studies have added objective parameters to this classification to improve it.[13-21]

In 2013, the European Society of Human Reproduction and Embryology (ESHRE) and the European Society for Gynaecological Endoscopy (ESGE) gave a new classification based primarily on anatomy of the female genital tract. Anomalies were classified progressively according to the degree of the anatomical deviation. Co-existent cervical or vaginal anomalies are classified independently. This new classification is expected to be more precise and presentable and would provide the ease in deciding management protocol according to anomaly description.[22] Somayya M. Sadek et al has compared the performance of the classification sytems also.[23] Still any of these classification do not classify our anomaly. We would like to name it as a hemi cavity canalization defect. As it had a normal size and shaped uterus with 1 half of the uterine cavity being non cavitated. The fallopian tube also on that side was non-canalized and rudimentary.

CONCLUSION

Authors stress upon the rarity of this case. There has been no similar case reported in the literature till date, adding to the spectra of complex anomalies. This also emphasizes on the need for classifying the unclassified anomalies.

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