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Research Article

A RARE CASE OF OBSTRUCTIVE COMPLEX UTERINE ANAMOLY MASQUEREDING AS COMPLEX OVARIAN CYST

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Abstract

Mullerian duct anomalies, though rare, can be a treatable cause of pelvic pain and infertility. Various complex Mullerian duct anomalies may exist with combination of features of more than one class. 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. In this case, 16yr old female with normal secondary sexual characteristics presented with acute abdominal pain and primary amenorrhea. Patient was evaluated by sonography and MRI and diagnosed as complex uterine anomaly of unicornate uterus with cervical dysgenesis and non-communicating rudimentary horn with large complex cyst with ovarian origin. There should be an integrated clinico-radiological classification scheme and familiarity with rare and complex anomalies for appropriate diagnosis and management of complex Müllerian duct anomalies. Though many cases of complex Mullerian anomalies have been described so far, this presentation of functioning rudimentary horn separately lying free in peritoneal cavity hasn't been reported yet, adding to spectrum of complex anomalies.

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Keywords: Mullerian Duct Anomaly, Uterine Obstruction, Hematosalpinx, Ovarian Cyst Mimic

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1. Introduction

Developmental anomalies of the Mullerian duct system represent some of the most fascinating disorders that obstetricians and gynecologists encounter. The Mullerian ducts are the primordial anlage of the female reproductive tract. They differentiate to form the fallopian tubes, uterus, uterine cervix, and superior aspect of the vagina. A wide variety of malformations can occur when this system is disrupted[1]. They range from uterine and vaginal agenesis to duplication of the uterus and vagina to minor uterine cavity abnormalities. When more than 1 complete or incomplete defect coexists, they lead to formation of a complex anomaly[2]. Here we report a case of an unusual and unclassified complex uterine anomaly and its management

CASE REPORT

A 16-year-old female presented with complaints of acute abdominal pain since 3 days. She had history of primary amenorrhea, chronic pelvic pain for 2 years aggravating cyclically associated with nausea and vomiting. General physical examination was unremarkable with normal secondary sexual characteristics. Per abdomen showed 2 separate palatable domino pelvic masses with right sided mass present about 20 weeks with firm in consistency, left sided mass present about 24 weeks variable in consistency and separately palpable. Pelvic examination revealed normal external genitalia with well-developed vagina, blind ending cervix with no external ostium. Patient had been invested many times, pelvic sonography done outside with inconclusive reports. She was operated for cervical canalization twice prior but with failed attempts

INVESTIGATIONS

Patient underwent sonography in radiology department which demonstrated distended uterus oriented more towards right. Right ovary normal. Cervix visualized with no endocervical canal. Left sided complex mass comprising solid & cystic components likely arising from ovary which is suggestive of endometrioma.

MRI pelvis showed presence of 2 asymmetric uterine horns, large on right side with hematometra of 11x6 cm, endometrial thickness of 7mm and right ovary of 3x1.7cm. Left uterine rudimentary horn with 4.1x3.2cm showing no communication. The left solid cystic lesion with internal septations of 10.6x4cm arising from left ovary likely complete ovarian cyst. Cervical well developed about 4x3 cm with no endocervical canal and not communicating with endometrial cavity, vagina measuring 7x2cm.

On basis of MRI findings, a diagnosis of unicornuate uterus with cervical dysgenesis, non-functioning, non communicating rudimentary horn with left sided complex ovarian cyst. (Figure 1)

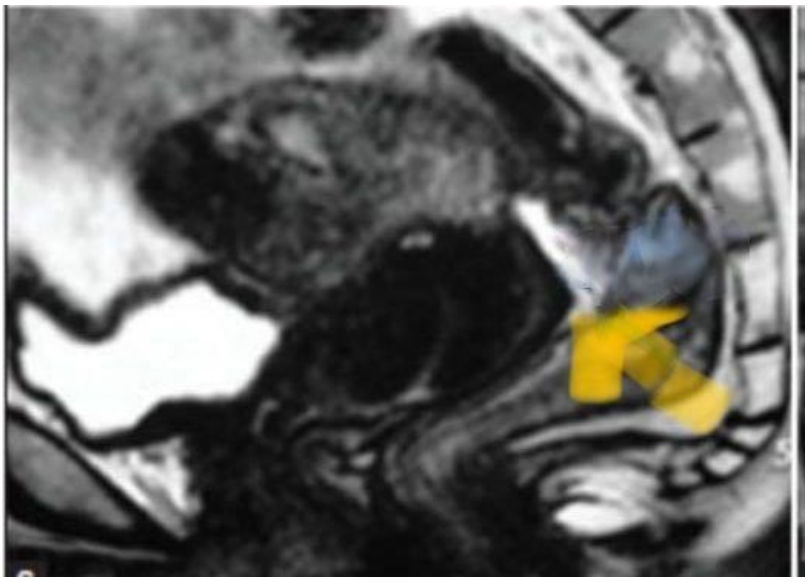

Figure 1. Sagittal T2W image showing well-formed cervix (arrow) with no communication between endometrial cavity and endocervical canal.

The patient was taken up for hysterolaparoscopy. The cervix was well developed but the hysteroscope could not be negotiated beyond the isthmus, consistent with cervical dysgenesis. Dense adhesions were found in the pelvic cavity, likely due to retrograde

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menstruation and Stage IV endometriosis. The omentum was adherent to the uterus and the pouch of Douglas was obliterated. Right hemiuterus (distended – 10x8cms hematomata) with cervical dysgenesis and a blind left non-functioning rudimentary horn with large hematosalphinx (10x10cm) noted. The left nonfunctioning horn with its fallopian tube dilated, thick, congested, oedematous lying separated from parent horn and dense adhesion with left ovary radiologically masquerading as complex ovarian cyst. (Figure 2, 3)

Figure 2. Hysterectomy specimen with right hemiuterus with hematometra (10x8cms) and cervical dygenesis,
left non communicating horn with large hematosaphinx (10*10cm) separated from parent horn

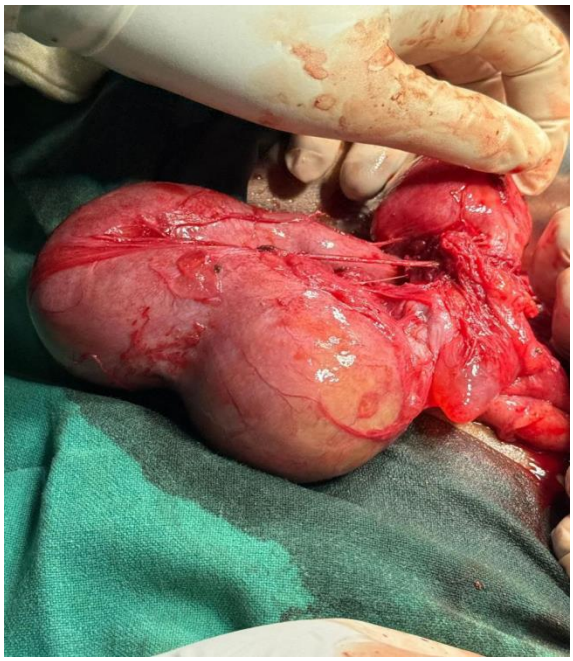
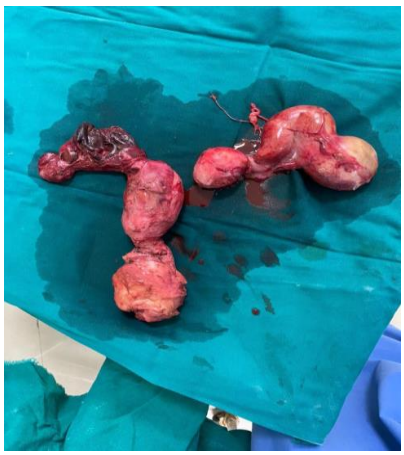


Figure 3. Left rudimentary horn with hematosaphinx with only mesosalphingeal attachment and separately lying from parent horn(adehesions between hematosaphinx and left ovary released).

Due to delayed presentation of the patient, advanced endometriosis, dense pelvic adhesions and a failed attempt to



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recanalize/reconstruct cervix, a decision for hysterectomy was taken after obtaining patient's consent. Both uterine horns with fallopian tubes were removed and adhesiolysis was done. Vaginal vault sutured and patent vagina for sexual function preserved. There were no intra or postoperative complications

DISCUSSION:

Genetic causes of Mullerian duct anomalies are complicated and uncommon. Inheritance patterns can be autosomal dominant, autosomal recessive, and X-linked disorders. Mullerian anomalies can be part of a multiple malformation syndrome[1,2]. Studies have estimated that Mullerian duct anomalies (MDA) can affect between 4 percent and nearly 7 percent of the female population[3,4]. Though rare, may present in different ways from infancy to young adulthood, with mucocolpos, hematocolpos, hematometra, primary amenorrhea, pelvic pain, infertility, or repeated pregnancy loss.

An understanding of the differences between these uterovaginal anomalies is important in understanding the respective clinical manifestations, different treatment regimens and prognosis. Many classifications of uterine anomalies developed including the Buttram and Gibbons and the American Fertility Society (AFS) classification[5]. Though these classifications are important in the treatment of symptoms arising from obstruction or deformity, it is important to realize that these classification systems serve merely as a framework and not all anomalies will fit completely into one of these categories. It is even more important to accurately describe the different components of the complex anomalies so that appropriate management can be planned.

The modified AFS classification by Rock and Adam[6] embraces a broader collection of uterine and vaginal anomalies without conflicting observations or over simplicity encountered in other classifications. This classification correlates anatomic anomalies with embryologic arrests, classifying uterovaginal anomalies as dysgenesis disorders or vertical or lateral fusion defects. Anomalies are further subcategorized into obstructive or nonobstructive forms, since their treatment differs. Class IV of this classification is a useful addition, to include unusual configurations or combination of defects, since genital tract aberrations do not necessarily follow any defined and consistent pattern.

A unicornuate uterus with a rudimentary horn is the most uncommon uterine anomaly, representing approximately 20% of all MDA. A cavitated, non-communicating rudimentary horn in a unicornuate uterus represents approximately 4.4% of all MDA. The treatment of this anomaly is surgical resection of the rudimentary horn.

Congenital cervical anomalies, including agenesis and dysgenesis are even rarer, with less than 200 cases of cervical agenesis being reported till date[7].

Many authors have recommended hysterectomy as an initial procedure for a patient with cervical dysgenesis/ agenesis with a functioning uterine corpus. This eliminates needless suffering from associated problems of endometriosis, sepsis and multiple surgeries. Considering the small potential for pregnancy, some authors advocate procedures of recanalisation or reconstruction of cervix, especially in cases of cervical dysgenesis[7].

Although many of these anomalies may be initially diagnosed at hysterosalpingography or sonography, further imaging by MRI is often required for definite diagnosis and elaboration of secondary findings. MRI has a reported accuracy of up to 100% in evaluating MDA. Lately 3D-US is highly accurate for diagnosing uterine malformations, having a good level of agreement with MRI .3D -USG imaging provides clear delineation of internal and external uterine anatomy[8]. Complex anomalies and secondary findings of endometriosis along with renal anomalies should also be looked for.

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The present case is unique, comprising of combination of two rare anomalies, a unicornuate uterus with cervical dysgenesis and a blind non cavitated noncommunicating rudimentary horn with patent fallopian tube. Embryologically this anamoly is due to more than one developmental defect, explained by

1. complete lack of fusion of both Müllerian ducts with no anatomical continuity
2. Incomplete development of right hemiuterus with cervical dysgenesis and left partially cavitated rudimentary horn.

This anomaly can be assigned to class IV of modified AFS classification by Rock and Adam. The review of literature did not reveal a similar case, though many authors have emphasized the possibility of complex anomalies in a patient with features of more than one class. In the present case, the diagnosis was delayed due to complex nature of the anomaly and absence of an integrated clinico-radiological classification scheme for specific diagnosis of such anomalies.

CONCLUSION:

To conclude, various complex MDA may exist with combined features of more than one class. An integrated clinical and radiological classification and familiarity with rare and complex MDA is essential. Early surgery offered to the patient may reduce patients suffering, help restore a patent outflow tract and may preserve fertility in some cases.

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