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CASE REPORT

A Rare Case of Recurrent Vaginal Wall Abscess in Communicating Bicornuate Uterus with Renal Agenesis

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ABSTRACT

Clinical presentation of müllerian anomalies vary widely. We are reporting here a case of müllerian anomaly, had suffered from secondary infertility and purulent vaginal discharge for last two years. After thorough clinical examination and systematic investigations, her proper diagnosis of communicating bicornuate uterus (H shaped) with partial agenesis of left cervix and total agenesis of right cervix and right kidney was made and patency of right tube was restored, so that not only her symptoms were resolved but she had successful pregnancy too.

Keywords: Bicornuate uterus, Recurrent vaginal abscess, Renal agenesis.

INTRODUCTION

Müllerian duct anomalies are although not very rare (overall incidence 2-3%)[1] but have a diverse spectrum of presentation. We are presenting a rare case of recurrent vaginal wall abscess with communicating bicornuate uterus associated with absent right kidney and secondary infertility who later on had a successful pregnancy.

CASE REPORT

A 24-year-old female reported in our OPD as a case of recurrent vaginal abscess with complaint of heaviness in lower abdomen associated with pain and vaginal discharge, greenish yellow in color for last 2 years with history of secondary infertility.

She had a previous history of dilatation and evacuation for blighted ovum of 7 weeks along with vaginal abscess, one and half years back in some other institute. After 2 weeks of termination, repeat curettage was done for retained products of conception. At that time, though the USG report was showing lateral vaginal wall abscess with bicornuate uterus, and patient was giving history of drainage of abscess but no documents were available with her for exact procedure.

She attended menarche at the age of 13 years. Her menstrual cycles were regular with average flow associated with mild dysmenorrhea. Her general condition was fair, and vitals were stable. On per abdominal examination, her abdomen was soft without any swelling or tenderness.

On Per Speculum Exam

On left side, anterior lip of cervix was seen with ill-defined posterior lip. On right side, cervix was not visualized separately. Greenish yellow, thick, mucoid discharge was present coming from right side of vaginal vault with a small cystic swelling at right anterolateral vaginal wall.

On Per Vaginal Exam

Cervix was soft and poorly formed. Uterus was 6 weeks in size, deviated to right side with irregular contour. Uterus was firm and mobile, bilateral fornices were free and nontender.

INVESTIGATIONS

Ultrasonography

It revealed absent right kidney and normal left kidney, two uterine horn with evidence of heterogenous collection in right uterine horn cervix and vagina.

Bilateral ovaries were normal.

CT Scan

It is confirmed that the diagnosis showing absent right kidney with bicornuate bicollis uterus with thick-walled abscess in endocervical and endometrial canal on right side continuing up to lateral vaginal wall with normal left kidney.

In view of abscess and suspected müllerian malformation, her examination under anesthesia, incision and drainage with diagnostic hysterolaparoscopy was planned. Diagnostic laparoscopy was suggestive for bicornuate uterus with bilateral fallopian tubes and ovaries were normal. Chromopertubation was not done due to presence of active infection (Fig. 1).

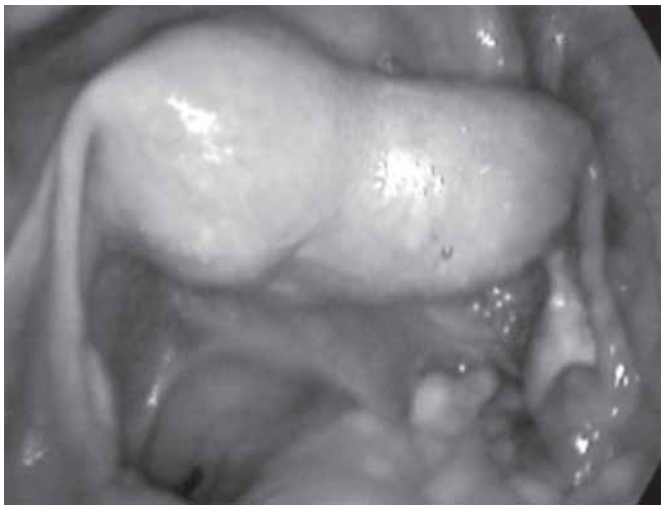

Fig. 1: Diagnostic laparoscopy showing bicornuate uterus with normal bilateral tubes and ovaries

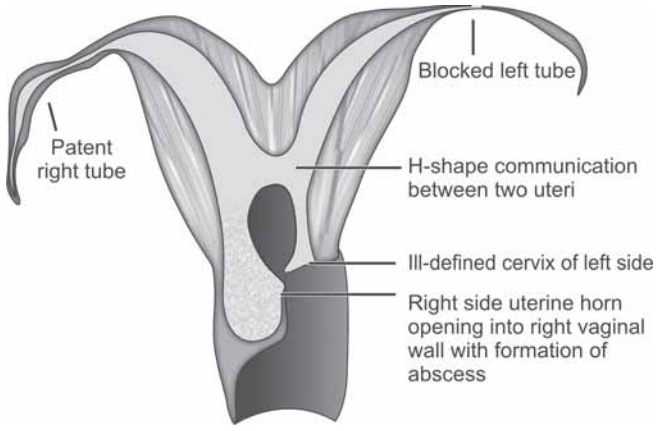

Fig. 2: Müllerian duct anomaly with abscess

Diagnostic Hysteroscopy

Bilateral cornual openings were visualized separately when hysteroscope was passed through cervix. To confirm the communication with vaginal wall abscess, a cannulation wire was passed through the cervix, which came out through the right side from abscess cavity to vaginal wall. No distinct cervix was seen on right side.

Abscess cavity was drained, marsupialization was done to maintain the patency, pus and abscess wall, was sent for culture which came positive for *E.Coli* sensitive for Amikacin. After 6 weeks of diagnostic laparoscopy and antibiotic therapy, fluroscopic sinogram with hysterosalpingography was done which shown a horizontal communication between two uterine cavities, i.e.'H'-shaped uterus, patent right fallopian tube and blocked left-sided tube.

Patient was under follow-up and advised contraception for 6 weeks. Later on patient conceived spontaneously after 3 months and presently has completed 28 weeks of gestation.

Lateral vaginal wall abscess was due to partial obstruction of right-sided horn.

DISCUSSION

Final diagnosis of this case was recurrent vaginal wall abscess (i.e. pyocolpos and pyometra) in communicating bicornuate uterus with right sided cervical, renal agenesis, and partial agenesis of left cervix. Lateral vaginal wall abscess was due to partial obstruction of right-sided horn (Fig. 2). Successful pregnancy is rare to happen in such cases.

A bicornuate uterus results from partial nonunion of the müllerian ducts, the central myometrium may extend to the level of the internal cervical os (bicornuate unicollis) or external cervical os (bicornuate bicollis). The latter is distinguished from didelphys uterus because it demonstrates some degree of fusion between two horns, while in classic didelphys uterus two horns and cervices are separated completely. In addition, the horns of the bicornuate uteri are not fully developed; typically, they are smaller than those of didelphys uteri. The syndrome of obstructed hemivagina and ipsilateral renal anomaly was initially reported in 1922 and is known as Herlyn-Werner-Wunderlich syndrome or, more recently, by the acronym OHVIRA.[1-3] The OHVIRA syndrome classically occurs in the setting of uterine didelphys or, more rarely, a septate uterus. Renal agenesis is the most commonly reported urologic anomaly in OHVIRA syndrome, although other malformations including renal duplication and multicystic dysplastic kidney have also been described,[1] similar cases are reported by few other authors in literature.[1,2,4,5] The true incidence of an obstructive müllerian anomaly is not known because only those anomalies that result in pregnancy wastage or cause symptoms are generally reported. One should examine the urogenital system when a genital anomaly is identified and vice versa.[3] Renal agenesis is the most commonly reported anomaly occurring in 67% of cases, other anomalies include ectopic kidney, horseshoe kidney, renal dysplasia, and duplicated collecting systems when skeletal defects are present. The anomaly is referred to a MURCS association (Müllerian aplasia, renal aplasia, cervicothoracic and somatic dysplasia). Renal agenesis and/or ectopy occur in 88% of MURCS patients.[3] Women with müllerian anomalies may be asymptomatic or may suffer from a wide range of symptoms, which can manifest themselves at various stages of life, from childhood to senescence; before pregnancy, during pregnancy, and after. Symptoms cover a wide spectrum, varying with the type of anomaly and the age of the woman. There may be cyclic or noncyclic pelvic pain, dyspareunia, pelvic mass, purulent discharge and infertility. The main reasons for frequent diagnostic delay and/or inappropriate surgery are: (i) Not considering the malformation as a cause of the patient's clinical symptoms and (ii) not considering the embryological origin of the different constituent elements of the genitourinary tract (Acién, 1992).[3,4]

CONCLUSIONS

Knowledge of the genitourinary embryology best explains the malformative anatomic findings of uterus, and to make a proper diagnosis of complex malformations, careful clinical examination, ultrasound, hysterosalpingography and IV pyelogram, as well as CAT, MRI and appropiate laparoscopic or laparo-

tomic observation may be needed. One should think about it and act on the malformation according to embryological deductions, trying to correct only what is necessary to avoid present and likely clinical manifestations, because systematic supervision, proper diagnosis and management can lead to favorable outcome in these cases.[2]

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