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CASE REPORT

Missed Diagnosed Bicornuate Unicollis Uterus Presenting as Acute Abdomen

Diagnostique Manqué D' Utérus Bicorne Unicollis Présentant Comme Abdomen Aigu

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ABSTRACT

Uterus bicornuate unicollis is one of the various congenital abnormalities of the female genital tract caused by partial fusion of the mullerian ducts on both sides. A case of acute abdomen resulting from missed diagnosed bicornuate unicollis uterus in a 42-year infertile women was presented. The ultrasonographic diagnosis of twisted complex left adnexial cyst was made prior to expioratory laparotomy. However, findings at laparotomy revealed a non- communicating bicornuate unicollis uterus with damaged right tube (hyrosalpinx) and normal ovaries and left tube. A blind-ended rudimentary left sided uterine horn was exetsed with the ovary spared and the right sided hydrosalpinx disconnected using chromic 1. Patient was however counselled for *in vitro* fertilization. **WAJM 2014; 33(3): 222–224.**

Keywords: Uterus bicornuate unicollis, congenital abnormalities, acute abdomen.

RÉSUMÉ

L'utérus bicorne unicollis est l'une des diverses anomalies congénitales de l'appareil génital féminin causées par une fusion partielle des conduits de Müller sur les deux côtés. Un cas d'abdomen aigu résultant de diagnostic manqué d'utérus bicorne chez une femme infertile de 42 ans a été décri. Le diagnostic échographique de kyste tordu annexiel gauche complexe a été fait avant la laparotomie exploratrice. Cependant, les résultats de la laparotomie ont révélé un uterus bicorne non-communicant, avec un tube droit endommagé (hyrosalpinx), un tube gauche et des ovaires normaux. Une extrémité rudimentaire de la corne gauche de l'utérus a été excisée avec l'ovaire épargné et l'hydrosalpinx droit a été déconnectée à l'aide de chromique 1. La patiente a toutefois été conseillée pour une fécondation in vitro. **WAJM 2014; 33(3): 222–224.**

Mots clés: Utérus bicorne unicollis , anomalies congénitales, abdomen aigu

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Abbreviations: **AFS,** American Fertility Society; **EVS,** Endovaginal Sonography; **HSG,** Hysterosalpngography; **USS,** Ultrasound.

INTRODUCTION

Developmental anomalies of the müllerian duct system represent some of the most fascinating disorders that obstetricians and gynecologists encounter. The müllerian ducts are the primordial anlage of the female reproductive tract.[1] They differentiate to form the fallopian tubes, uterus, the uterine cervix, and the 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.[1-3] Because of the wide variation in clinical presentations, müllerian duct anomalies may be difficult to diagnose. After an accurate diagnosis is rendered, many treatment options exist, and they are usually tailored to the specific müllerian anomaly.[4,5]

Bicornuate uterus is a type of uterine duplication anomaly resulting from partial nonfusion of the müllerian ducts.[6] The central myometrium may extend to the level of the internal cervical os (bicornuate unicollis) or external cervical os (bicornuate bicollis). It can be categorized as class IV Mullerian duct anomaly according to the American Fertility Society (AFS) classification scheme.[7]

The true prevalence is unknown because the anomalies usually are discovered in patients presenting with infertility.[5] However the overall incidence of mullerian duct anomalies is estimated to occur in 0.1–0.5% of women.[1,6] Bicornuate uteri are thought to represent approximately 10–39 % of Mullerian duct anomalies.[8]

Patients with müllerian duct anomalies are known to have a higher incidence of infertility, repeated first-trimester spontaneous abortions, fetal intrauterine growth retardation, fetal malposition, preterm labor, and retained placenta.[6] The rarity of this mullerian duct anomaly presenting as acute abdomen prompted the report of this case.

CASE REPORT

A 42-year old nulligravid woman who presented to the out-patient clinic of Omolola Specialist Hospital, Ilorin, Kwara State, Nigeria with a 10 years history of infertility and 2 years history of recurrent cyclical mild lower abdominal pain. There was positive history of sexually transmitted infection a year prior to presentation. There were no significant findings on physical examination at presentation.

Hysterosalpingography showed normal sized uterus with non-demonstration of the left tube and right sided hydrosalpinx. The 2D endovaginal ultrasound examination of the pelvis revealed a left sided complex adnexeal mass which measures 5cm by 6cm. Hormonal profile results were normal. She was however counseled for therapeutic laparoscopy and was referred to the University of Ilorin Teaching Hospital, North Central Nigeria.

At about two months later she represented with a 12-hour history of sudden onset of severe lower abdominal pain and vomiting of no relieving or aggravating factors which coincided with the onset of her menstrual flow. A repeat pelvic 2D endovaginal ultrasound was ordered which was suggestive of features of torsion of the left ovarian cyst. She was however prepared for exploratory laparotomy.

Operative findings (Fig. 1) revealed a non-communicating bicornuate unicollis uterus with hydrosalpinx of the right tube, the ovaries and the left tube were normal. A subserous fibroid nodule at the fundal pole of each uteri. The right uterus (larger (6cm x 5cm)) has a communication with the cervix while the left (smaller 3cm x 3cm)) do not. A blind-ended rudimentary left sided uterine horn was excised with the ovary spared and the right sided hydrosalpinx disconnected using chromic 1. The excised left uterus was cut open and was observed to be filled with altered blood with clots and was sent for histology. Histology result showed uterine tissue with heamatometria. She was subsequently counseled for *in vitro* Fertilization and was discharged home after seven days of admission.

Fig.1: Operative findings: A non-communicating bicornuate uterus with diseased right tube (hydrosalpinx) and normal left tube and ovaries. A subserous fibroid nodule at the fundal pole of each uteri.

DISCUSSION

The preferred methods of imaging uterine anomalies are ultrasound, hysterosalpingogram (HSG) or MRI.[6] The external uterine contour is concave or heart shaped, and the uterine horns are widely divergent, the fundal cleft is typically more than 1cm deep and the inter-cornual distance is widened in bicornuate uterus.[5] While HSG is of utmost importance in anatomical diagnosis of bicornuate uterus especially in non-pregnant patient, ultrasonography provides a very safe and non-invasive imaging modality for pregnant and non pregnant patient.[6]

The diagnosis of bicornuate uterus can be suggested by hystero-salpngography (HSG) or ultrasound (USS) though with very low sensitivity and specificity.[6] On the contrary, MRI has consistently demonstrated very low false-negative and false-positive rates for evaluation of uterine anomalies.[4] Pellerito et al also noted that MRI had the added advantage of detecting other incidental abnormalities, including a dermoid and submucosal leiomyoma, found on endo-vaginal sonography to be indeterminate and nonvisualized, respectively.[9]

On HSG and USS, a common finding is separation of the uterine cavity into right and left compartments with intercornual distance greater than 4cm. Although not a specific finding, the angle between the horns of the bicornuate uterus is usually more than 105° and the differentiation between the bicornuate and septate uterus is often difficult due to the outer uterine contour not being visible.[5] On MRI scans, the findings of bicornuate uterus are: two uterine cavities are seen with normal endometrial and myometrial width and ratio, myometrial characteristics of the

septum are accurately diagnosed, a deep (> 1 cm) fundal cleft in the outer uterine contour and an inter-cornual distance of more than 4 cm.[6]

In this case report. The diagnosis of mullerian duct anomaly was not entertained because of the reports of ultrasonography and HSG. However, this is not surprising as the tests have their limitations. Nicolini *et al* found that transabdominal 2D US failed to visualize the uterine cavity adequately in as many as 35% of patients although it adequately imaged the uterine fundus in 90% of patients.[10] Studies have also found 2D transvaginal sonography to be a highly effective means of diagnosis, with 75-100% sensitivity and up to 95% specificity. Positive predictive value was higher with 3D scanning than with 2D scanning (100% vs 50%, respectively).[10,11] Unfortunately, 3D US which is highly sensitive (up to 100%) and specific (up to 100%) in helping diagnose major müllerian anomalies[11] was not available in the centre.

HSG techniques did not provide diagnoses with high degrees of confidence due to existence of a large overlap between the subtypes of mullerian duct abnormalities when comparing uterine cavitary configuration, intercornual distance, and intercornual angle.[6,12] Therefore, anomalies incidentally discovered on HSG are referred for further evaluation using MRI or US since their diagnostic precision is better.[6] Therefore, HSG findings in this report need to be cautiously interpreted.

MRI has consistently demonstrated high sensitivity and specificity for evaluation of uterine anomalies. Pellerito et al found MRI capable of helping correctly diagnose 24 of 24 anomalies (100% accuracy), compared to 11 of 12 anomalies (92%) detected on endovaginal sonography (EVS).[12] For anomalies requiring surgery (unicornuate or bicornuate uteri), MRI demonstrated 100% sensitivity and specificity, compared to 67% sensitivity and 100% specificity for EVS.[4] For non-surgical lesions, both MRI and EVS had 100% sensitivity and specificity.[4] MRI was not ordered in this case because non-availability and cost has hampered its use in the evaluation of uterine anomalies in the centre.

In conclusion, Müllerian anomalies are a morphologically diverse group of developmental disorders that involve the internal female reproductive tract. Establishing an accurate diagnosis using 3D ultrasound scan and MRI is essential for planning treatment and management strategies.

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