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BICORNUATE UNICOLLIS UTERINE MÜLLERIAN ANOMALY WITH UNILATERAL HAEMATOMETRA IN A TEN YEAR OLD: A CASE REPORT

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SUMMARY

Acute abdominal pain is a common complaint in the paediatric age group. Gastroenteritis and appendicitis are the common non-surgical and surgical conditions respectively. Haematometra due to a non-communicating horn of a bicornuate uterus is a rare presentation for acute abdomen in this age group. This is a case of a ten year nine month old female who presented with an acute abdomen for a period of two days. Physical examination revealed right iliac fossa tenderness. Pelvic ultrasound showed a bicornuate unicollis uterine Müllerian anomaly with unilateral haematometra. Magnetic Resonance Imaging (MRI) confirmed the ultrasonographic findings. A successful hemi-hysterectomy was done with resolution of symptoms.

INTRODUCTION

The female genital tract arises from the formation and fusion of Müllerian ducts during the embryogenic period, giving rise to the fallopian tubes, uterus, cervix and the upper two-thirds of the vagina. Failure of formation of the Müllerian ducts results in uterine agenesis/hypoplasia or a unicornuate uterus. Conversely, failure of fusion results in a bicornuate or didelphys uterus. To complete the development of these organs, the septum in between the ducts gets resorbed. Defects in the resorption stage result in a septate or arcuate uterus (1, 2).

A bicornuate uterus is characterised by partial non-fusion of the Müllerian duct. This results in a central myometrium that may extend to the level of the internal cervical os (bicornuate unicollis) or external os (bicornuate bicollis) (2).

CASE REPORT

A ten year nine month old female presented to the emergency department with a two day history of severe sharp abdominal pain worse on the right iliac fossa. It was associated with two episodes of vomiting.

However, there was no associated hotness of body, urinary symptoms or bowel changes. There was no history of abnormal vaginal discharge. Her last normal menstrual period was three days prior to admission. Menarche was five months from presentation. She had had two cycles since then, light flow, lasting five days. She has no history of sexual activity or contraceptive use.

On physical examination, her vital signs were within normal range. Right iliac fossa tenderness with no rebound tenderness was elicited on abdominal examination. Vaginal exam showed, normal external genitalia with an intact hymen, hence speculum and digital examination was differed.

An abdomino-pelvic ultrasound showed a bi-cornuate unicollis uterine Mullerian anomaly with a unilateral haematometra with a volume of 5.83 cc. A Magnetic Resonance Imaging (MRI) done confirmed the ultrasonographic finding and further showed that the cornu were separated by a septum measuring 17mm, the left cornu was normal in size continuing to the cervix and vagina and a mass effect on the superior aspect of the bladder, displacing the right ureter laterally was demonstrated in the right cornu (Image A). Full blood count was normal range,

except for slight neutrophilia. Urea, electrolytes and creatinine parameters were within normal ranges.

A decision for hysteroscopy and a laparoscopic hemi-hysterectomy was made. Informed consent was given by the patient's mother. The laparoscopic approach was unsuccessful, and open hemi-hysterectomy was done successfully (Image A, B, C and D).

Image A

Ultrasound image of the bicornuate uterus behind the urinary bladder



Image B

MRI image of the saggital section of the pelvis showing an enlarged horn, with a second horn communicating with the cervix



Image B

Intra operative - enlarged right horn with a blind ended fallopian tube



Image D

Hemi-hysterectomy specimen - right horn with fallopian tube in a kidney dish



Patient did well post operatively and was discharged on the second post operative day. On the one week follow up visit, the patient was doing well with no complaints. The wound was clean and healing well, with no abdominal tenderness.

DISCUSSION

Evidence suggests that, about 2% of the general population have major uterine anomalies (3). A bicornuate uterus results from incomplete fusion of the mullerian ducts at the level of the fundus, creating two separate cavities with a common lower segment and cervix. A patient with bicornuate uterus may present with one or two cervixes (4). The depth of the midline cleft varies with the severity of the fusion abnormality (3).

Females with a bicornuate uterus have a higher prevalence of pregnancy loss but are asymptomatic, and, because these women have fewer reproductive problems than do women with other anomalies, the pathologic condition may go undiagnosed until cesarean delivery, or sometimes as an incidental finding on imaging (4). Typically, however, uterus didelphys with an obstruction becomes symptomatic at menarche, generating cyclic pelvic pain (dysmenorrhea) that coincides with regular menses from the unobstructed side (4). This obstruction can cause retrograde menstrual flow, which explains the increased prevalence of endometriosis and pelvic adhesions in affected patients. Due to a similar obstruction, this is how our patient presented.

The risks to future obstetric career of a bicornuate uterus include: recurrent early pregnancy losses, preterm deliveries and cervical incompetence (3). Uterine rupture is another rare presentation in primigravidas with a malformed uterus. The incidence of pregnancy in rudimentary horn is rare, about one in 40,000 pregnancies (5).

Treatment for a bicornuate uterus depends on the presentation. In case of recurrent pregnancy losses, unification procedures may be done, the procedure of choice being the Strassman abdominal metroplasty. Cervical cerclage may be done in those presenting with cervical incompetence. In this case however, a

hemi-hysterectomy was considered as the best course of treatment.

This case emphasises the importance of comprehensive clinical examination, importance of imaging and a high index of suspicion in the diagnosis and management of haematometra due to a bicornuate uterus in the pediatric age group. In conclusion, there should be a high index of suspicion for a congenital uterine anomaly in the female paediatric age group presenting with an acute abdomen.

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