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CASE REPORT

A Case of Rare Mullerian Anomaly—Functional Rudimentary Uterine Horn: As an Unusual Cause of Unilateral Dysmenorrhea in Postmenarchal Adolescent Girl

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ABSTRACT

An 18-year-old girl presented with progressively increasing dysmenorrhea. A pelvic mass was suspected which on exploratory laparotomy was found to be a rudimentary horn (hematometra, hematosalpinx). We report a case of unicornuate uterus with functioning rudimentary horn to highlight that mullerian duct anomalies should be considered in the differential diagnosis of severe dysmenorrhea even in normally menstruating girls. Unicornuate uterus with a rudimentary horn is susceptible to many gynecologic and obstetric complications. Hematometra, chronic pelvic pain, endometriosis, infertility are some of the complaints in women with unicornuate uterus.

Keywords: Rudimentary horn, Unicornuate uterus, Dysmenorrhea, Hematometra.

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INTRODUCTION

The unicornuate uterus is caused by nondevelopment of one mullerian duct. The condition usually is associated with various degrees of rudimentary horn connected to the unicornuate uterus when one of the ducts develop only partially. Various series show the rarity of such an anomaly, it has been reported to be of incidence of 0.06%. It is generally considered that the presence of a noncommunicating cavitatory rudimentary horn (Buttram and Gibbons class IIA, B) carries a poor reproductive prognosis and increases the risks of endometriosis and cornual pregnancy. Mullerian anomalies are commonly associated with renal, spinal and cloacal anomalies. There is particular association of unilateral renal agenesis and unicornis uterus.[1] The prevalence of dysmenorrhea is estimated to be around 25% of all women and up to 90% of adolescents.[2]

CASE REPORT

An 18-year-old girl presented in the gynecological outpatient department with onset of dull aching pain in the left iliac fossa since 3 years. The patient had spasmodic progressive dysmenorrhea since menarche, i.e. from 14 years. The patient complained of feeling a mass in the lower abdomen since past 1 year. Her menstrual cycles were regular since menarche with moderate flow but with progressively increasing dysmenorrhea. Patient had imperforate anus at birth for which she had undergone a series of corrective surgical procedures during her neonatal period. Patient had been on antispasmodic pain killers for her dysmenorrhea. When the patient presented in the gynec outpatient department, a thorough examination revealed a tender cystic mass palpable in the left iliac fossa 10×6 cm.

All routine examinations revealed no obvious abnormality. Beta hCG was negative. USG suggested differential diagnoses of left hematosalpinx , hematometra and bicornuate uterus, with nonvisualized left kidney. CT scan shows ill-defined multiloculated lesion in pelvic cavity and both ovary are not seen separately with collection in endometrial cavity (Fig. 1).

To confirm the diagnosis, magnetic resonance imaging (MRI) was performed, which showed a unicornuate uterus with left-sided cavitary rudimentary horn with left hematometra and hematosalpinx and left renal agenesis (Fig. 2). Intravenous pyelography also confirmed left renal agenesis. On urology consultation, cystoscopy was performed, which revealed non-visualization of the left ureteric orifice with normal right ureteric orifice and bladder mucosa. A diagnostic laparoscopy was performed which revealed a right unicornuate uterus with patent right fallopian tube and normal right ovary. Right unicornuate uterus was connected to rudimentary horn on the left side with hematometra and hematosalpinx invoving the left horn and tube. Endometriotic lesions were seen over the peritoneum mainly on the left side with partial obliteration of the cul-de-sac with involvement of sigmoid colon by adhesions which were flimsy.

Due to some infrastructural problems, exploratory laparotomy was performed over the preference for operative laparoscopy. On laparotomy, the unicornuate uterus was

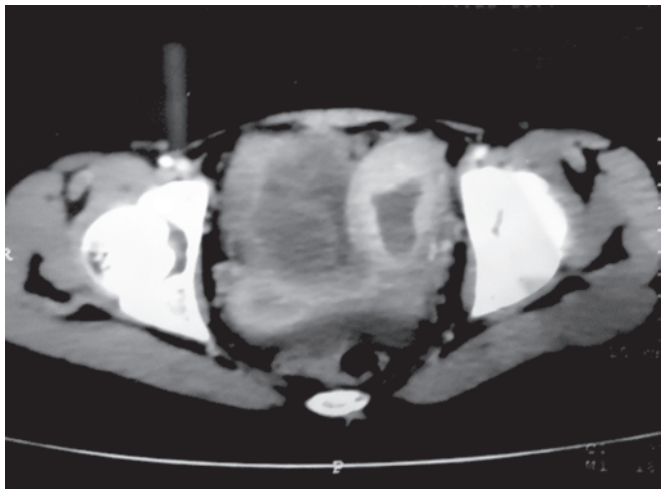

Fig. 1: CT scan: Ill-defined multiloculated lesion in pelvic cavity and both ovaries are not seen separately

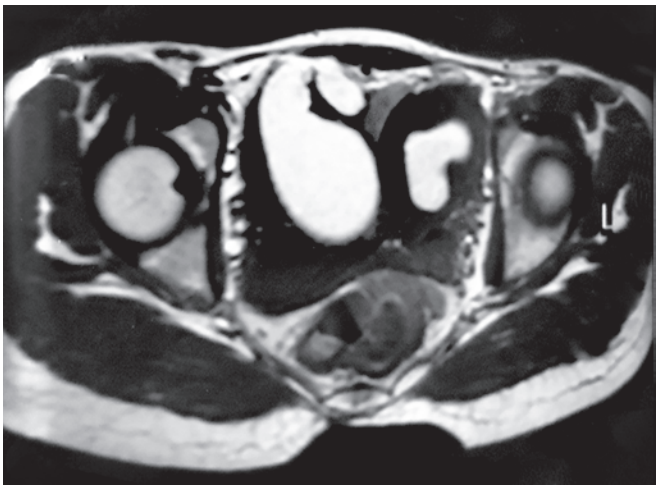

Fig. 2: MRI shows unicornuate uterus attached to large left cornu with collection in endometrial cavity of left cornu and hyperintense tubular structure noted seems to be fallopian tube attached to left side of left cornu s/o hematosalpinx

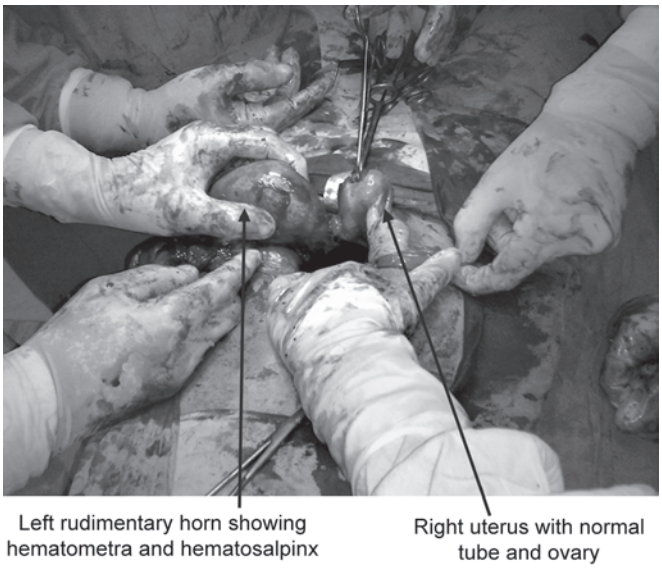

Fig. 3: Intraoperative photograph showing right unicornuate uterus with normal tube and ovary attached to left noncommunicating rudimentary horn showing hematometra and hematosalpinx

connected to rudimentary horn on the left side with hematometra and hematosalpinx invoving the left horn and tube (Fig. 3). On incising the left-sided horn of the uterus, anchovy sauce-like material came out. The left-sided rudimentary horn was found to be noncommunicating with the main uterine cavity. Excision of the left-sided cornu and left salpingo-oophorectomy was performed maintaining proper hemostasis. Endometriotic lesions over the peritoneum were coagulated and adhesions involving the bowel and cul-de-sac were lysed. Patient had an uneventful postoperative recovery. The patient had attended our outpatient department for follow-up and had no complaint of dysmenorrhea.

DISCUSSION

Unicornuate uterus with a rudimentary horn is a rare type of mullerian duct malformation, with a reported incidence of 0.06%.[3] In 83% of the cases, the rudimentary horn is noncommunicating.[4] The rudimentary horn may consist of a functional endometrial cavity or it may be a small solid lump of uterine muscle with no functional endometrium. Mullerian anomalies are commonly associated with absence or gross malformation of the renal tract. It is important to keep an index of suspicion in high-risk groups of uterine or mullerian anomalies regarding other common spinal, cloacal and renal anomalies.[1]

A unicornuate uterus causes few symptoms and it is usually discovered by chance or as a result of pregnancy complications. But, in our case, the patient presented with unilateral dysmenorrhea. When dysmenorrhea is with rudimentary horn, it is usually because of the obstruction to the outflow of menstrual blood. Other causes of unilateral dysmenorrhea may be endometriosis with unilateral distribution or a small leiomyoma at the uterotubal junction. One-sided spasmodic dysmenorrhea in a young girl should always raise the suspicion of uterine malformation, and every effort should be made to exclude the condition by conducting relevant investigations. The diagnosis of mullerian abnormalities can be made via USG. However, MRI is more specific for the evaluation of presence or absence of a functional endometrium. But, the gold standard to diagnose mullerian anomaly is diagnostic laparoscopy.

It is generally considered that the presence of a noncommunicating cavitary rudimentary horn carries increased risks of endometriosis and cornual pregnancy. Removal of a noncommunicating horn is recommended to reduce the risk of recurrent or *de novo* endometriosis.[5] It is difficult to truly estimate the incidence of such complications as the data available are in the form of case reports and they usually present as surgical emergencies. To avoid serious complications in the future, early diagnosis and excision of the rudimentary horn are of utmost importance. If facility and expertise are available, laparoscopic excision should be offered especially to the young girls as it is less morbid and cosmetically more acceptable.

CONCLUSION

Unilateral dysmenorrhea, although unusual, might present as acute abdomen. It should be thoroughly evaluated especially in adolescents with relevant investigations keeping in mind the rare possibility of mullerian anomaly. Early diagnosis and prompt treatment is recommended in these cases to avoid future gynecological and obstetrical complications. The right step at the right moment aided with maximal suspicion and investigation and minimal access surgery is the preferred mode of management here.

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