

Non-communicating Rudimentary Horn with Haematometra Manifesting as Adnexal Mass: A Case Report

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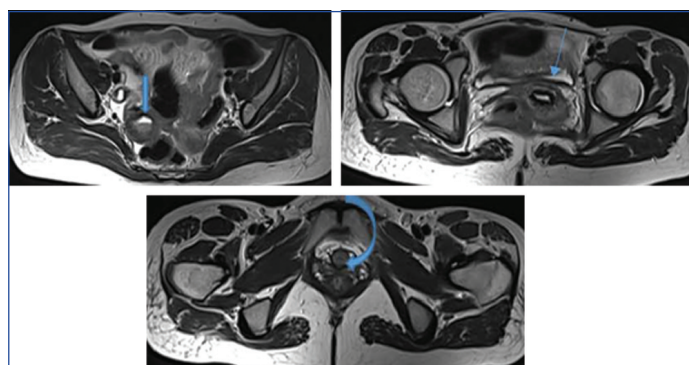
ABSTRACT

Female reproductive organs other than ovaries are formed from the müllerian ducts which develop into fallopian ducts, uterus, and the upper two third of vagina. Any defective formation and non-fusion of müllerian ducts leads to multiple anomalies with varying combinations with variable presentations in these affected women. Anomalies like rudimentary horn without non-functional endometrium are asymptomatic and incidentally diagnosed while undergoing imaging studies like ultrasound and magnetic resonance imaging of pelvis. Anomalies of rudimentary horn with functional endometrium, patients may have dysmenorrhoea, abnormal bleeding at the time of menarche, ectopic pregnancy in the rudimentary horn, and endometriosis. The patient might experience a delayed onset of presentation, and some women may end up with life-threatening conditions such as ruptured ectopic pregnancy in the rudimentary horn. A 35-year-old female with a history of dysmenorrhoea for 15 years and recent aggravated pain, presented for imaging of the pelvis. Right adnexal lesion was suggested during the ultrasound imaging, and the Magnetic Resonance Imaging (MRI) pelvis suggested a rudimentary horn on the right-side with blood collection within the horn, causing haematometra. Laparoscopic removal of the rudimentary horn was done, and the presence of functional endometrium and myometrial tissue was confirmed in the histopathological examination. Early detection of these müllerian anomalies will help us prevent the morbidity and mortality associated with them.

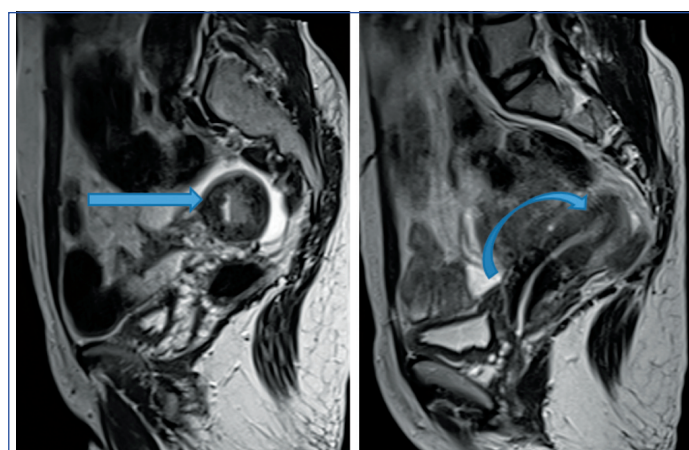
Keywords: Congenital uterine malformation, Dysmenorrhoea, müllerian anomalies, Pelvic pain, Unicornuate uterus

CASE REPORT

A 35-year-old woman came with a chief complaint of post-menstrual lower abdominal pain since 6 months, associated with fever and vomiting. She had a history of dysmenorrhoea for the last 15 years, regular cycle (3/30 cycle), normal flow, no clots. Mild pain during menstruation and post-menstruation. She was P2, L1, A1, (para 2, live one, abortion 1) with a history of normal vaginal delivery. Abdominal pain was located mainly in the right iliac fossa region and radiated to the right lower limb and thigh. The last childbirth was 13 years ago. The patient was being treated for hypothyroidism with tablet Thyroxine 12.5 mcg for the last five years. No other comorbidities like pulmonary tuberculosis, hypertension, and diabetes were noted. A previous surgical history of puerperal sterilisation was noted. Physical examination revealed tenderness in the right iliac and supra pubic region. Per vaginal examination showed bulky uterus and tenderness at right lateral fornix. Ultrasonography (USG) pelvis shows a thick-walled fluid-filled, oval-shaped structure in the right adnexa and is diagnosed as the right para ovarian cystic lesion. Both ovaries were identified and appeared unremarkable. Uterus and both kidneys were unremarkable. MRI abdomen showed a normal uterus and bilateral ovaries with the presence of oval shaped structure measuring 3.3×3.2 cm with a central area of blood collection with Susceptibility Weighted Imaging (SWI) blooming within right adnexa and raising the possibility of a rudimentary horn with haematometra [Table/Fig-1-4]. The right fallopian tube was mildly dilated [Table/Fig-4]. Both kidneys were normal in size, shape, and position [Table/Fig-5]. Vagina appeared normal. Subsequently, laparoscopic removal of the right horn with the fallopian tube was performed. Histopathological examination confirmed the presence of endometrial and myometrial tissues in the right adnexal lesion and confirmed it as a non-communicating uterine horn with haematometra. The patient is on routine follow-up and there has been marked symptomatic relief on subsequent visits.



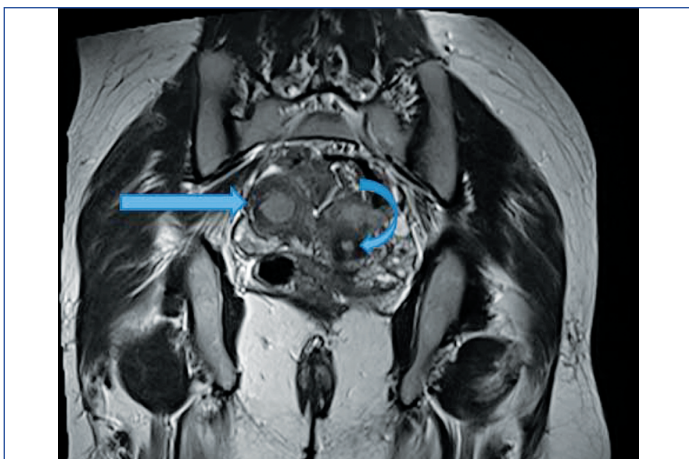
[Table/Fig-1]: Axial T2 w images show thick walled blood filled with fluid level in right rudimentary horn (arrow), normal cervix (thin arrow) and vagina (curved arrow).



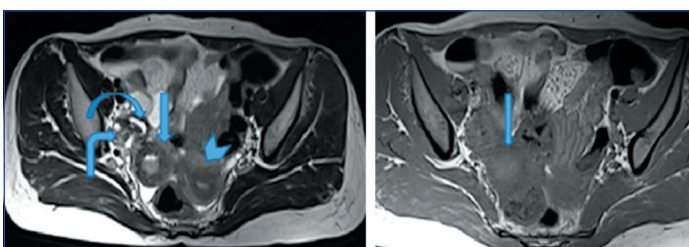
[Table/Fig-2]: T2 sagittal images show a normal uterus and endometrial cavity (curved arrow) and right rudimentary horn with fluid level and surrounding free fluid (bold arrow).

DISCUSSION

In embryonic life, the female reproductive system is formed from two müllerian ducts and develops into fallopian ducts, uterus, cervix



[Table/Fig-3]: Coronal T2 W images show blood-filled right rudimentary horn (Bold arrow) and uterus with endometrial cavity (Curved arrow), part of right and left ovaries.



[Table/Fig-4]: Axial T2/T1 W/SWI images show thick-walled blood-filled rudimentary horn (arrow), dilated right fallopian tube (curved arrow), right ovary (bended arrow) and normal uterus (arrow head).



[Table/Fig-5]: Coronal HASTE shows bilateral normal kidneys and normal liver and spleen.

and vagina. The proximal part of the Müllerian ducts is unfused and forms two fallopian tubes proximal end forms the fimbria of fallopian tubes. The fused part of the Müllerian ducts forms the uterus, and cervix. The distal end of fused mullerian ducts forms the upper two third of vagina. The distal third of vagina is formed by bilateral sinovaginal bulbs, which in turn arise from the urogenital sinus [1,2]. Failure of formation, and fusion of the Mullerian ducts lead to multiple Mullerian anomalies. Based on the American fertility association, Mullerian anomalies have been divided into 7 classes [3] and our case falls under Class II: Unicornuate uterus:

- Class I: Hypoplasia/uterine hypoplasia. (Mayer Rokitansky Kuster Hauser syndrome);

- Class II: Unicornuate uterus;
- Class III: Uterus didelphys;
- Class IV: Bicornuate uterus;
- Class V: Septate uterus;
- Class VI: Arcuate uterus;
- Class VII: T-shaped uterus resulting from the exposure to diethylstilbestrol in foetal life.

Incidence of the rudimentary non-communicating horn with a functional endometrial cavity is very rare and often challenging to diagnose because of the variations in clinical features and clinical presentation. These clinical manifestations may be clinically asymptomatic, often missed during a routine gynaecological examination, or manifest as dysmenorrhoea, or infertility and amenorrhoea. These mullerian anomalies, especially the unicornuate uterus usually associated with genitourinary tract, gastrointestinal tract abnormalities, since all three genital, urinary tract, and gastrointestinal tract drain into common opening till 5th week of embryonic development [4,5]. Renal hypoplasia or aplasia is observed in approximately 15% of patients with a unicornuate uterus, with ipsilateral renal anomalies present in 40% of these cases, typically corresponding to the side of rudimentary horn [4,6]. In addition, a unicornuate uterus with ovarian endometriosis has also been reported [7].

In this case report, the uterus appeared normal in morphology, size, and shape, which was confirmed during the laparoscopy procedure. Histopathological examination revealed myometrial and endometrial tissue in the resected rudimentary horn. The patient also had normal vaginal delivery indicating the normal morphological, functional, and reproductive nature of uterus. The presence of a rudimentary horn with normal functional uterus is a rare association. Even though the patient had long history of dysmenorrhoea, this condition was picked up relatively later during the imaging studies for abdominal pain. Apart from this, concomitant renal anomalies were also ruled out in this patient. No associated renal, vertebral, and anal anomalies were detected. In this case report, the presence of rudimentary horn with the delayed onset of presentation and complications was diagnosed by imaging studies and treated successfully.

Garapati J et al., presented a case of 28-year-old pregnant women, who had a history of one previous normal vaginal delivery, and reported with premature rupture of the amniotic membrane and abdominal pain. She underwent an emergency caesarean section and delivered a baby weighing 2900 gm without any complications. The patient had a unicornuate uterus with non communicating rudimentary horn. This emphasises the significance of early detection of uterine anomalies to prevent maternal and foetal morbidities and mortality, and suggests the necessity for additional research and large-scale studies on these complex pregnancies [8].

Rackow BW et al., studied the reproductive performance of women with Mullerian anomalies. They concluded that even though Mullerian duct anomalies are uncommon, these are associated with normal and abnormal reproductive outcomes such as recurrent miscarriage, preterm labour, early rupture of the amniotic membrane, and stillbirth. Each variant of Mullerian duct abnormalities has its reproductive implications, and most of these anomalies need surgical intervention. Management of Mullerian anomalies with infertility remains controversial [9].

Singh P et al., reported a case of unruptured pregnancy in a non-communicating uterine horn with torsion of the gravid horn and ipsilateral tube, ovary, with moderate ascites, which was diagnosed by MRI and later confirmed surgically and histopathologically. This patient had two previous full-term normal vaginal deliveries [10]. Pregnancy in a non-communicating rudimentary horn is extremely rare with an incidence of 1 in 76000-150000 pregnancies, and usually terminates with rupture of the rudimentary horn [10].

Behrens M et al., reported a case of infected haematometra in a rudimentary non-communicating horn misdiagnosed as a pelvic mass. A 20-year-old woman, who had presented with acute abdominal pain and was found to have 8 cm oval-shaped cystic right adnexal lesion, was diagnosed with pelvic inflammatory disease. Since, there was no improvement in symptoms and reduction in the size of the adnexal lesion after the course of antibiotics, an infected non-communicating rudimentary horn was suspected. Robotic-assisted laparoscopic removal of this adnexal lesion was performed, and the nature of the non-communicating rudimentary horn was confirmed in the histopathological examination [11]. This patient is more similar to this case report, initially misdiagnosed and later confirmed to be a case of Müllerian anomalies.

CONCLUSION(S)

Varying radiological studies are helpful in the evaluation of a rudimentary non-communicating horn. USG computed tomography, MRI, sonohysterography and hysterosalpingogram could be useful in identifying this lesion. These mullerian anomalies are uncommon and difficult to diagnose early, and present with delayed complications. MRI is the most sensitive and specific method in the evaluation of this condition. Early intervention prevents mortality and morbidity of both the mother and foetus and prevents complications.

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