

# Dysmenorrhea due to a rare müllerian anomaly

M Agarwal, A Das, AS Singh

Department of Obstetrics and Gynecology, North Eastern Indira Gandhi Regional Institute of Health and Medical Sciences Shillong, India

## Abstract

Müllerian duct anomalies may produce reproductive failure like abortion and preterm birth, or obstetric problems like malpresentation, retained placenta, etc., or they may be asymptomatic. Unicornuate uterus with a noncommunicating functional rudimentary horn is a type of müllerian anomaly that results in obstruction to menstrual blood flow, leading to endometriosis and dysmenorrhea. Though the majority of cases of dysmenorrhea in adolescents are primary in nature and require only reassurance and symptomatic management, it is important to be aware of rare causes such as müllerian anomalies so that these cases can be properly managed. Hence, we present this case report, with interesting illustrations, so as to increase awareness regarding these anomalies.

**Key words:** Dysmenorrhea, müllerian anomaly, unicornuate uterus

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## Introduction

Unicornuate uterus with a rudimentary horn is a rare type of müllerian duct malformation and is the result of defective fusion of the malformed duct with the contralateral duct.<sup>[1]</sup> The incidence of unicornuate uterus, although not precisely known, is estimated at 1/1000 women.<sup>[2]</sup> A noncommunicating rudimentary horn with a functional endometrial cavity is rare and can be associated with many complications throughout a woman's reproductive life, beginning from menarche when hormonal stimulation may gradually activate the endometrium of the rudimentary horn. The resulting obstruction may cause retrograde expulsion of menstrual debris and, as per Sampson's theory, lead to endometriosis and infertility. Unicornuate uterus is found in 0.4% of women of reproductive age and it is associated with endometriosis in up to 40% of cases.<sup>[2,3]</sup>

We report one such case that developed endometriosis and had to undergo surgical removal of the horn for resolution of symptoms.

## Case Report

An 18-year-old unmarried girl attended our outpatient

### Address for correspondence:

Dr. Manika Agarwal,  
Department of Obstetrics and Gynecology, North Eastern Indira Gandhi Regional Institute of Health and Medical Sciences, (NEIGRIHMS), Shillong, India.  
E-mail: drmanika89@yahoo.com

department with complaints of severe pain in the lower abdomen during her menses for the last 6 months. Apart from severe dysmenorrhea there was no other menstrual abnormality. Her vitals and per abdominal examination findings were normal. Ultrasonography of the abdomen suggested the possibility of unicornuate uterus with right-sided hematosalpinx and hematometra; also, the right kidney was not visualized. Her blood reports were normal, except for CA 125 which was 228.3 IU/ml. Intravenous pyelography revealed a nonvisualized right kidney. On the urologist's advice, cystoscopy was done; this showed a normal left ureteric orifice and bladder mucosa, with nonvisualization of the right ureteral orifice. To confirm the diagnosis, magnetic resonance imaging (MRI) was done. This showed a unicornuate uterus with a functioning cavitory rudimentary horn on the right side, with right-sided hematometra and hematosalpinx. Lobulated, organized blood was found in the right side of the pelvis and there was a hemorrhagic cyst in the right ovary and also right renal agenesis. The noncommunicating horn was considered to be the cause of the patient's severe

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dysmenorrhea due to intracavitary retention of menstrual effluent and retrograde menstrual flow. The patient was then taken for laparotomy. Intraoperatively, right-sided hematometra and hematosalpinx were seen, [Figure 1] as also a hemorrhagic right ovarian cyst [Figure 2]. On incising the right-sided hematometra, anchovy sauce-like material came out. On trying to negotiate the cavity with a Hegar dilator, it was found to be noncommunicating with the main uterine cavity. Excision of the right cornu and tube, with right-sided ovariectomy, was done. The left tube and ovary, along with the uterus, were preserved. The abdominal cavity was cleaned and closed after achieving complete hemostasis. The excised specimen was sent for histopathological examination. The postoperative period was uneventful [Figure 3].

Histopathology revealed a rudimentary horn with endometrial and myometrial tissue. The right tube showed chronic salpingitis, with areas of hemosiderin-laden macrophages. The ovary had features suggestive of a simple hemorrhagic follicular cyst. The patient is being followed up in the outpatient department and is now relieved of her dysmenorrhea.

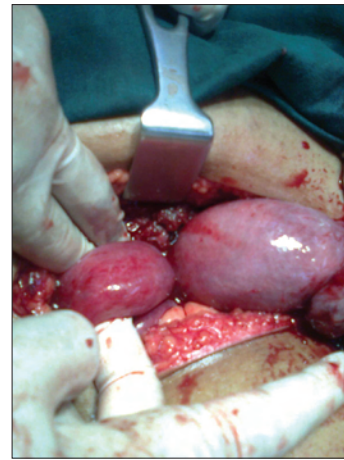
## Discussion

Unicornuate uterus with a noncommunicating functional rudimentary horn may lead to various gynecological and obstetric complications. Coexisting urinary tract abnormalities are frequent in these patients and are dominated by unilateral renal agenesis, homolateral to the side of the rudimentary horn,<sup>[2]</sup> as was seen in our case also. Though most cases of dysmenorrhea are of the primary type and can be managed by reassurance and some supportive treatment, routine ultrasonography needs to be done to rule out any organic pathology. If required, further investigation (e.g., MRI) can be done to confirm the diagnosis. MRI is excellent for depicting the uterine morphology in cases of unicornuate uterus with rudimentary horn noncommunicating with the main cavity and distended by hematometra and associated hematosalpinx.

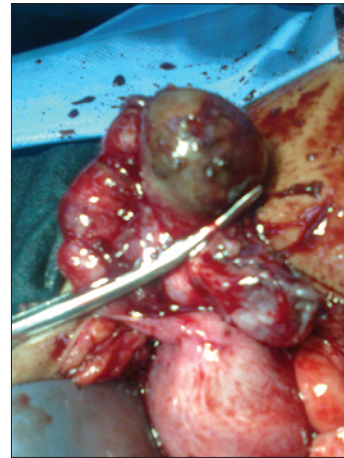
Based on various data, radical management is recommended in case of a noncommunicating cavitary rudimentary horn so as to avoid the risk of endometriosis.<sup>[4]</sup> In our case, obstruction of the outflow tract of the rudimentary functional horn, with consequent menstrual collection, led to dysmenorrhea. Retrograde flow of the menstrual collection gave rise to endometriosis and its consequences, for which surgery was needed.

## Conclusions

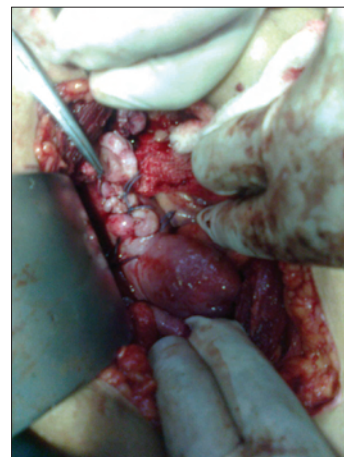
The triad of dysmenorrhea beginning at menarche, increasing severity of dysmenorrhea over time, and a



**Figure 1:** Hematometra and hematosalpinx



**Figure 2:** Right ovarian cyst



**Figure 3:** Post resection

unilateral pelvic mass is strong evidence of a congenital müllerian duct anomaly. Such a strong association requires thorough investigation including, when necessary, MRI.

Radical surgical removal of the anomaly is still the mainstay of management of such an anomaly. The basic objective of surgical resection of a functional noncommunicating rudimentary horn is pain relief and prevention of endometriosis and its consequences.

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