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## RESEARCH ARTICLE

### UNICORNUATE UTERUS WITH NONCOMMUNICATING FUNCTIONAL HORN : A CASE REPORT

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#### ABSTRACT

Mullerian duct anomalies represent a group of congenital anomalies that result from arrested development, or incomplete fusion of the mesonephric ducts. The incidence of congenital mullerian anomalies is 1/200 to 1/600. The incidence of rudimentary horn is very rare (1:100,000). Approximately 75% of such horns do not communicate with the normal hemiuterus. These uterine anomalies are either diagnosed incidentally or the patient may present with obstetrical or gynecological problems. Vaginal obstruction is associated with perivaginal mass, pain, and endometriosis, but cyclic menstrual flow may be present because of the normally functioning opposite side. This anomaly is usually associated with ipsilateral renal agenesis (67%) or ipsilateral pelvic kidney. Laparotomy has been the standard approach for such cases. We report this case in hope of expanding the knowledge of a rare occurrence.

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## INTRODUCTION

Mullerian duct anomalies represent a group of congenital anomalies that result from arrested development, or incomplete fusion of the mesonephric ducts [Raga, 1997]. Depending on the incompleteness of fusion, there is a wide variety of malformations. The American Fertility Society (AFS) suggested a specific classification for uterine anomalies [American Society for Reproductive Medicine, 1988]. The incidence of congenital uterine anomalies in fertile population is 1/200 to 1/600. The incidence of rudimentary horn is very rare (1:100,000). Approximately 75% of such horns do not communicate with the normal hemiuterus. A unicornuate uterus is a rare uterine malformation with an incidence of 2.5–13% [Chakravarti, 2003]. Incomplete fusion of the two Mullerian ducts may lead to a septate or bicornuate uterus; failure of Mullerian tube formation causes aplasia or atresia of one side, resulting in a unicornuate uterus. Incomplete atresia of a Mullerian duct leads to a rudimentary horn which is broadly connected or connected through streak tissue with the unicornuate uterus [Falcone et al., 1997].

Non-communicating and functional rudimentary horn can cause severe pain in abdomen due to accumulation of the blood causing its distension. Also, pregnancies in these rudimentary horns may be diagnosed late, with a 70% risk of uterine rupture occurring before 20 weeks of gestation accompanied by potentially massive and life-threatening intraperitoneal haemorrhage [Kadir, 1996]. These uterine anomalies are either diagnosed incidentally or the patient may present with obstetrical or gynecological problems. Vaginal obstruction is associated with perivaginal mass, pain, and endometriosis, but cyclic menstrual flow may be present because of the normally functioning opposite side. This anomaly is usually associated with ipsilateral renal agenesis (67%) or ipsilateral pelvic kidney.

## CASE REPORT

A 29 year old patient, married since 7 years, nulligravida, came to GMCH Nagpur with presenting complaints of abdominal pain during menses since 10 years (severe dysmenorrhoea), abdominal distension since 5 years. There is no h/o bladder and bowel complaints. The dysmenorrhoea was not relieved by analgesics and antispasmodic medications and OCP's. Patient gives h/o operated for some abdominal lump in 2009, details of which are not available.

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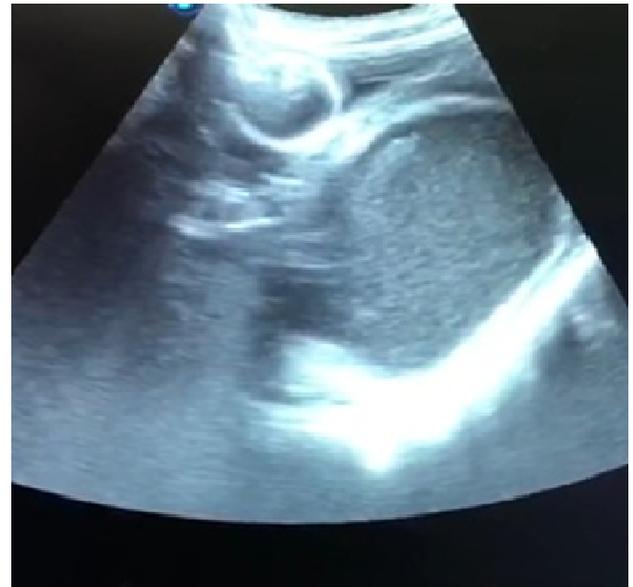
On Examination-Her vitals, general, physical and systemic examination were normal. On Abdominal examination- 32 weeks mass palpated, solid in consistency, no side to side mobility seen, nontender. On per speculum examination-a single cervix was seen with normal vagina. The cervix was deviated to left side. Pelvic ultrasound revealed a large irregular complex mass in the right side of abdomen with multiple cystic and solid components, 25 × 8 cm right ovarian mass (Fig. 1). The uterus was deviated to left with absent right kidney. Left ovary was normal. The right ovary could not be identified. She was posted for diagnostic laparoscopy, SOS exploratory laparotomy. On Diagnostic Laparoscopy, a unicornuate left hemi- uterus with a non-communicating right horn was noted with a fibrous band in between. Laparoscopy showed a left unicornuate uterus with normal adnexa, a large right non-communicating horn. As complete anatomy of the non-communicating horn could not be delineated, decision of laparotomy was taken.

On exploratory laparotomy, there was single kidney on left side. On laparotomy, the uterine anomaly was confirmed. The left uterine horn was small, with normal left tube and ovary and round ligament attachment. The right horn had no communication with surrounding structures except for round ligament attachment which was cut; anterior fold of peritoneum was cut and pushed down. (Fig. 2) The right horn was separated circumferentially and feeding vessels with fibrous band identified at the base, clamp applied at the base, pedicle cut and transfixed with vicryl. Cut section of the right horn showed that the cavity was filled with dark chocolate coloured fluid with myohyperplasia. (Fig 3, 4) Histological study reported it as a cavitated adenomyotic rudiment. She had an uneventful post-operative recovery. She has been pain free after surgery.

## DISCUSSION

Unicornuate uterus with functional non-communicating horn is a rare condition but is associated with various gynecological and reproductive morbidities. The recent ESHRE/ESGE classification system for mullerian anomalies suggests Class U0 as normal uteri, Class U 1 as dysmorphic uteri, Class U2 septate, Class U3 bicorporeal, Class U4 hemi-uterus, Class U5 atrophic and Class U6 unclassified. As the literature demonstrates, early diagnosis is of great importance in order to avoid consecutive damage of the reproductive system and further painful complications [Jayasinghe, 2005; Dimitrova, 1997; Kriplani, 2001]. To get to the correct diagnosis, the patient's history is crucial: increasingly painful periods, dyspareunia and sterility are common symptoms [Chakravarti, 2003; Atmaca, 2005].

Our patient complained of increasing post-menstrual pain, which, in retrospective, we consider to be due to the increasing size of the rudimentary horn. If in patients presenting with infertility, hysterosalpingography shows that the uterus is deviated to one side and there is unilateral tubal block, this condition should be strongly suspected. Pregnancy in non-communicating rudimentary horn is a rare form of ectopic gestation and its incidence is between 1/ 100,000 to 1/140,000 pregnancies. It occurs following transperitoneal migration of sperm or zygote. Variable thickness of rudimentary horn musculature, dysfunctional endometrium and poor distensibility of the myometrium lead to rupture of the rudimentary horn.



**Fig. 1. Ultrasonography showing rudimentary horn with hematocolpos**



**Fig. 2. Intra-operative picture showing normal size uterus with left sided ovary and fallopian tube. It also shows Right sided functional noncommunicating horn with hematocolpos inside**



**Fig 3: Chocolate coloured fluid being hematometra drained out.**

This complication is usually seen in the 2nd trimester and can be a life threatening condition for the mother resulting from hemoperitoneum and hemorrhagic shock. It is difficult to diagnose preoperatively and in the literature, only 5% of rudimentary horn pregnancies were diagnosed preoperatively and the remaining were found unexpectedly at laparotomy. If the pregnancy occurs in the semi-uterus of this malformation, it is associated with increased incidence of abortion, preterm labor and malpresentations. These patients also have high incidence of cesarean deliveries. To obtain the proper diagnosis, endovaginal grey-scale sonography is mandatory. With regard to imaging techniques, magnetic resonance imaging (MRI) is the gold standard for the diagnosis of Mullerian abnormalities, although three-dimensional sonography is able to achieve comparable results. Both these techniques provide us with a detailed structure of the anatomical anomaly, hence enabling a proper plan with regard to the surgical approach. These patients should be managed by expert surgeon because wrong excision of normal horn can be a big problem for the future life of patient. Laparotomy has been the standard approach for such cases. But with emerging skills in minimally invasive surgery, laparoscopy has become the first choice. If undetected, a unicornuate with a non-communicating cavitated horn is associated with a high index of complications. The fact that hematometra, hematosalpinx or even endometriosis are consequences of retrograde menstruation has already been described. The reason our patient did not develop endometriosis and only showed hematometra could be explained by the existence of a hypoplastic right tube that prevented retrograde menstruation. In the present case, the procedure was effective in resolving the pelvic pain and dysmenorrhea and avoided the risk of endometriosis.

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