

# A Comparative Analysis of Cryptomenorrhea in Patients with Vaginal Obstruction Vis a Vis Patients with Rudimentary non Communicating Horn of Uterus with Functional Endometrium: A Retrospective Study.

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## ABSTRACT

**Background:** To analyse the clinical features, diagnostic challenge and treatment options in patients presenting with pain abdomen and cryptomenorrhea in patients with vaginal obstructive lesions in comparison with those having normal vagina but having non communicating rudimentary horn with functional endometrium. **Methods:** The clinical details of patients presenting with cryptomenorrhea to our institute was studied. The data collected was from a period of 7 years; from March 2010 to April 2017. The clinical details were collected from cases sheets of the respective cases. **Result:** A total of 6 cases were studied. Among the 6 patients, all were within the age group of 13-20 years. 3 patients had abnormality in the form of vaginal agenesis, transverse vaginal septum and imperforate hymen. All the 3 presented with amenorrhea and severe cyclical pain. The other 3 patients had rudimentary non communicating horn with functional endometrium and had different clinical presentations. One had severe dysmenorrhoea, one had severe cyclical pain with amenorrhea and the third patient had been operated earlier and presented with severe dysmenorrhoea with an incision site sinus with discharge of menstrual blood abdominally along with vaginal passage of menses. Since reflux of menstrual blood is seen in all cases, endometriotic ovary was found in 2 cases. One had only hematosalpinx. Two patients have been operated earlier for endometriotic cyst but the abnormality in the uterus or vagina was not detected, leading to reoccurrence of symptoms. **Conclusion:** Cryptomenorrhea or hidden menses due to congenital malformation in the uterus or vagina is a rare condition but can lead to distressing symptoms of dysmenorrhoea in young girls. It can also cause endometriosis due to retrograde menstruation. Improper evaluation can miss the proper diagnosis and patient may be operated only for endometriosis of ovary as a cause for dysmenorrhoea. Not identifying and treating the primary abnormality will lead to recurrence of symptoms and repeat surgeries in patients.

**Keywords:** Cryptomenorrhea.

## INTRODUCTION

Obstructive lesions of the vagina and non-communicating horn of the uterus with functional endometrium are both very rare congenital anomalies that can cause cryptomenorrhea and lead to distressing cyclical pain in young girls. Though, obstructive lesions of vagina may be diagnosed early on examination but its treatment may be difficult as they may be associated with varying cervical abnormalities such as cervical agenesis and atresia. On the other hand, rudimentary non communicating horn may be viewed on imaging studies and

sometimes peroperatively when patient has been operated only for co-existing endometriotic cyst leaving the primary abnormality behind. Thus our study aims to highlight the clinical features, diagnostic dilemma and treatment undertaken in these rare cases.

## MATERIALS AND METHODS

This study was conducted in a tertiary care teaching hospital. 6 cases were diagnosed to have dysmenorrhea due to various congenital malformations in the uterus or vagina leading to cryptomenorrhea in a 7 year period. All these patients were surgically treated. These patients were divided into 2 groups Group 1(n=3) consisted of patients diagnosed to have rudimentary non communicating horn with functional endometrium. Group 2 (n=3) consisted of patients with congenital

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outflow tract disorders such as vaginal agenesis, transverse vaginal septum, and imperforate hymen. Age, presenting complaints, reproductive history, investigations, preoperative diagnosis and intraoperative findings were studied.

## RESULTS

Group 1 (Patients having rudimentary non communicating horn of uterus with functional endometrium; n=3): Treatment given were as follows – In the first patient USG, and MRI were suggestive of unicornuate uterus with functional contrary rudimentary horn and haemorrhagic cyst in the right ovary. Patient was taken for surgery and right endometriotic cyst and rudimentary horn were excised. Patient relieved of symptoms.

The second patient presented with dysmenorrhea and menstrual blood discharging sinus from the incision site of surgery done elsewhere for ovarian cyst (2 years back). On admission, USG and MRI were suggestive of endometriotic cyst. She was given 3

monthly injection of Luprolide which led to temporary cessation of menses and relief to the patient for 6 months. Following this when symptoms recurred, repeat MRI showed endometriotic cyst and patient was taken up for surgery. On laparotomy she was found to have enlarged rudimentary horn attached to the incision site through sinus tract. Both the rudimentary horn and sinus tract were excised and patient relieved of symptoms.

The third patient had a small cervix and small uterus on the right side with a large 7X8 cm mass left in the left side abdomen. On vaginal examination, MRI abdomen showed a small horn of the uterus on the right side and a large blood filled horn of the uterus on the left side of the abdomen. Patient was taken up for laparotomy and a small rudimentary horn was found on the right side of uterus and a large non communicating horn was seen on the left side and excised. Post operatively patient was relieved of cyclical pain. She was started on oestrogen and progesterone to develop other horn, however patient was lost to follow up.

**Table 1: Clinical Profile Of Patients In Group -1.**

Age	Marital Status	Obstetric History	Complaints	Past Surgical History	Associated Malformation	Genital Tract Malformation
18	Unmarried	Nulligravida	Dysmenorrhea	None	Right Kidney Absent	Rudimentary Non Communicating Horn Of Uterus With Functional Endometrium
20	Unmarried	Nulligravida	Dysmenorrhea With Monthly Discharge Of Menstrual Blood Through Sinus On Incision Site Of Previous Surgery	2 Years Back Surgery For Ovarian Cyst	Right Kidney Absent	Rudimentary Non Communicating Horn Of Uterus With Functional Endometrium
18	Married	Nulligravida	Amenorrhea With Cyclical Pain	None	Polydactyl , Atrial Secundum In Heart	Rudimentary Non Communicating Horn Of Uterus With Functional Endometrium

**Table 2: Clinical Profile Of Patients In Group 2**

Age	Marital Status	Obstetric History	Complaints	Past Surgical History	Associated Malformation	Genital Tract Malformation
18	Unmarried	Nulligravida	Amenorrhea With Cyclical Pain	2 Years Back Operated For Ovarian Cyst	None	Vaginal Agenesis With Cervical Agenesis
16	Unmarried	Nulligravida	Amenorrhea With Cyclical Pain	None	None	Transverse Vaginal Septum
13	Unmarried	Nulligravida	Amenorrhea With Cyclical Pain	None	None	Imperforate Hymen

Group 2 (Patients with congenital outflow tract disorder, n=3): Treatment given were as follows – The first patient was found to have absent vagina on examination. MRI showed normal uterus with endometriotic cyst. Patient was taken up for surgery. Laparotomy was done and endometriotic cyst was excised. Uterus was identified however the lower part was embedded in adhesion. Vaginally, vaginoplasty was done but cervix could not be identified. Post operatively, patient was given injection luprolide in view of severe pain due to endometriosis and was counselled for hysterectomy. One month later hysterectomy was done and was found to have a uterus without cervix. Patient did well post operatively.

The second patient was found to have normal uterus with transverse vaginal septum on examination and

ultrasound. Patient was taken up for surgery and transverse vaginal septum was excised via abdominoperineal approach. A Foleys catheter was inserted through the cervix into the uterus and kept for 2 weeks. Patient did well post operatively.

The third case was found to have imperforate hymen with hamatometra on examination and ultrasound. She was treated by giving cruciate incision on the imperforate hymen. On follow up the patient was having normal menses.

## DISCUSSION

Vaginal agenesis is a rare mullerian malformation. Sometimes it is associated with absent uterus as in case of Mayer-Rokitansky-Küster-Hauser syndrome

(MRKH) or in Androgen Insensitivity Syndrome (Testicular feminising syndrome). In these cases patients generally present with primary amenorrhea or in case they are married with difficulty in having intercourse. In a study by Oya Soylu, a retrospective analysis of case was done who had undergone McIndoe's vaginoplasty due to congenital vagina agenesis.<sup>[1]</sup> In this study mean age of the patient was 28 years and most of them presented with primary amenorrhea or inability to have sexual intercourse. In their study most of the patients had absent uterus and ovaries. In our study age of presentation was much earlier as patient had functional uterus leading to haematometra thus severe monthly pain.

Most of our patients on laparotomy were found to have endometriosis due to retrograde menstruation. M.L.Marota et al also reported endometriotic implants in the peritoneum of the uterovesical space and pouch of Douglas of 11 year old who presented with severe abdominal pain for 4 months.<sup>[2]</sup> On evaluation she was found to have uterus with uterocervical septum with hamatometra, hematocolpos on both sides. She was operated and surgical resection of uterocervical septum was done. Imperforate hymen is a common cause of cryptomenorrhea, presenting around puberty usually. Ayse Secil EK Suglu has reported a case of imperforate hymen in an 8 month old girl who presented with restlessness and intermittent fever of unknown etiology.<sup>[3]</sup> Ultrasonography and MRI revealed grade hydronephrosis with hematocolpos that was causing marked distension of the uterus and cervix.<sup>[4]</sup> The diagnosis of imperforate hymen was made under sedation during instrumentation for the procedure by inspection of the protuberant mass on retraction of the labia. The patient was taken into operating room and a simple cruciate incision was made on the hymen under general anaesthesia which resulted in drainage of 500ml of yellowish non bloody mucosal secretion from vagina. On follow up, there was gradual resolution of hydronephrosis. Meraa Erna Vlahovic had reported a case of 12.5 years old girl who presented to the gynae emergency with pseudoacute abdomen with an abdominal mass and on examination was found to have slight bulging imperforate hymen and was treated with Mercedes hymenal incision and 1.2L of dark thick blood was drained out.<sup>[4]</sup>

Obi O Ansiem reported a case of acute urinary retention secondary to imperforate hymen in a 14 years old Nigerian girl which was treated by a cruciate incision on the hymen.<sup>[5]</sup> Aslude et al have also reported two cases of hematocolpos in adolescent girls due to imperforate hymen.<sup>[6]</sup> Both of them presented with lower abdominal pain and urinary retention and were treated with hymenectomy.

Though imperforate hymen is easy to diagnose and treat vaginal abnormality as vaginal agenesis and transverse vaginal septum are more challenging to

treat. Low transverse vaginal septae may be resected vaginally but medium/high septae are often more complex requiring abdominoperineal resection via laparotomy. Cara E Williams et al have reported laparoscopic approach of abdominoperineal resection of high transverse vaginal septae in two adolescent girls presenting with obstructed menstruation due to high transverse vaginal septae.<sup>[7]</sup>

Post-operative stenosis or stricture of vagina following excision of transverse vaginal septum is a complex complication seen in many cases. Ridhima Gupta et al presented a case of 15 year old girl with unicornuate uterus and transverse vaginal septum (TVS).<sup>[8]</sup> The TVS was excised, however patient was unable to perform vaginal dilation post operatively, leading to stricture formation. She underwent multiple surgeries for excision of the stricture. The patient was eventually evaluated every day in the clinic until she was able to demonstrate successful vaginal dilation in the presence of clinician. Hence the author concluded that regular and intense vaginal dilation after TVS excision may decrease the need for reoperation due to recurrent stricture formation.

In the analysis of varied presentation of congenital vaginal obstruction, Zafar Nazir et al reported 26 cases in 8 years period out of which 6 were neonates and 18 adolescents and 2 adults.<sup>[9]</sup> Common presentation in the neonate were abdominal mass, neonatal sepsis and respiratory distress, whereas abdominal pain, voiding dysfunction and backaches were prevalent in adolescents. Adult presented with inability to consummate and infertility.

Lavanya Kamaiyon et al have reported seven girls with cervicovaginal agenesis and four with lower vaginal agenesis (aged 12-20years) who presented with painful cryptomenorrhea,<sup>[10]</sup> where a colon conduit was constructed for the egress of menstrual blood. The colon conduit was anastomosed to posterior uterine wall in the seven girls with cervicovaginal agenesis and to the distended upper vaginal pouch in the four girls with vaginal agenesis. Utero-colonic neovaginal anastomosis was performed only after excising a circular portion of the posterior myometrium to prevent stenosis. They found that colon conduit functioned effectively; however, patients must be cautioned against pregnancy if they have vaginal agenesis.

In the assessment of sexual functioning, mental health and life goals in women with vaginal agenesis. Elizabeth M Morgan et al found that sexual functioning results were highly variable.<sup>[11]</sup> Participants reported significant emotional reaction at diagnosis as well as anxiety about the disorders specifically its role in relationships. However, overall the group showed average level of mental health and self esteem.

Unicornuate uterus is a very rare uterine anomaly. Unicornuate uterus with rudimentary horn is extremely rare with an incidence of 1 in 1,00,000.

Rudimentary horn of uterus may be communicating or non-communicating type and may be further subdivided as those containing functional or non functional endometrium. Rudimentary horn with non communicating non functional endometrium may be asymptomatic and incidental findings on an imaging or during some surgery.

However rudimentary horn which is non communicating with functional endometrium may have menstrual blood collecting inside leading to severe monthly pain. In a case reported by Anjali Rani et al a 17 year old girl presented with severe dysmenorrhea for the past 3 years.<sup>[12]</sup> Ultrasonography revealed two separate uterine cavities with hematometra in right side cavity with right ovarian cyst. The patient underwent diagnostic laparoscopy followed by laparotomy where they found the non communicating rudimentary horn attached by a fibrous band to the uterus and this horn was excised, following which her symptoms reduced.

In a similar case Rusen Atmaca found that a 27 years old woman who presented with dysmenorrhea for the past 13 years had a 3X4cm homogenous mass in the right adnexa both on pervaginal examination and on ultrasound.<sup>[13]</sup> On laparoscopy they found it was a case of non communicating rudimentary horn suspended by fibrous band to the uterus on right side. They excised the band laparoscopically and removed it.

In a study of different clinical presentation of patients with unicornuate uterus with non communicating horn, P Geol et al have analysed 18 patients.<sup>[14]</sup> Out of which 3 teenagers presented with dysmenorrhea, pain in abdomen, hematometra and underwent excision of rudimentary horn with ipsilateral salpingectomy. They found 7 patients had ectopic pregnancy which was surgically managed. 8 patients had intrauterine pregnancy and were diagnosed to have unicornuate uterus with non communicating rudimentary horn at the time of Caesarean delivery.

## CONCLUSION

Cryptomenorrhea is a very distressing situation for woman. Congenital anomalies such as vaginal outlet obstructive disorders or non communicating rudimentary horn with functional endometrium are rare disorders which may lead to cryptomenorrhea. Thorough knowledge of such disorders and proper preoperative evaluation and on table assessment during surgery is very important for accurate diagnosis and treatment of such anomalies.

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