

The perplexing entity of rudimentary uterine horn

Ria Malik, A. G. Radhika, Alpana Singh, Gita Radhakrishnan, Rachna Aggarwal

Department of Obs and Gynae, University College of Medical Sciences (UCMS) and GTB Hospital, Delhi, India.
Email: raradhikaag@gmail.com

Received 15 September 2011; revised 19 October 2011; accepted 5 November 2011.

ABSTRACT

A rudimentary uterine horn may be responsible for intriguing presentations in different stages of life. These presentations often masquerade commoner gynaecologic disorders resulting in diagnostic and management challenges for the treating clinicians. Whereas many of these anomalies may be discovered during the investigative workup for cryptomenorrhoea, dysmenorrhoea and infertility, due to lack of symptoms especially in a parous woman, a large proportion remains undiagnosed. We report here, two interesting presentations of this benign entity resulting in significant morbidity. The first case report describes late activation of a rudimentary horn presenting as chronic pelvic pain. The second patient presented with a failed second trimester induction of labor (abortion) for fetal demise. Her examination and investigations suggested an abdominal pregnancy, yet, on laparotomy, a 15 week pregnancy within an accessory uterine horn was discovered.

Keywords: Rudimentary Uterine Horn; Chronic Pelvic Pain; Laproscopic Management; Abdominal Pregnancy; Ruptured Rudimentary Horn

1. INTRODUCTION

Mullerian duct malformations represent a wide spectrum of anomalies, resulting from varying degrees of defects in the development and fusion of the mesonephric ducts. We present here, two unusual presentations of unicornuate uterus with rudimentary horn.

2. CASE I

Ms S, 37 yrs, P3L3 presented in our out patient clinic with complaints of dull aching lower abdominal pain since past 4 yrs. Recent history of dysmenorrhoea was noted. Her many visits to different gynecologists and surgeons did not prove fruitful. No symptoms suggestive of pelvic infection, bladder or bowel pathology were noted. Her first two children were term normal vaginal deliveries, third was a caesarian section with tubal ligation for

placenta previa, five years ago. Her postoperative period was uneventful and she was not informed of anything unusual at discharge; no papers relevant to the surgery were available when she presented to us. Her menstrual cycles had been regular and no period of amenorrhea was reported. General physical examination was unremarkable except for a pfannenstiel caesarean scar. Speculum examination revealed normal cervix and vagina. On vaginal examination, uterus anteverted multiparous size. There was a 4 cm × 4 cm size, mobile, smooth and slightly tender right adnexal mass. Transvaginal sonography showed a well circumscribed thick walled complex mass 4 × 3 cms in right adnexa with partially echogenic contents. Left ovary was normal. Provisional diagnosis of chronic pelvic inflammatory disease (PID) or possibly an endometrioma was considered. Her persistent symptoms despite treatment for PID mandated laparoscopy. At laparoscopy, the right ovary was found normal, the ligated right fallopian tube was seen arising from a 3 cm × 3 cm smooth surfaced thick muscular mass attached at the right uterine cornu. The right round ligament was attached to this mass confirming it to be rudimentary uterine horn. 4 ml of dark altered blood was aspirated from this mass. The other uterine half was normal in shape size and contour, left ovary and tube (ligated) were normal (**Figure 1**).

3. CASE II

25 yr old Ms. B, primigravida was referred by a practitioner for hysterotomy following failed attempt at second trimester termination of a nonviable pregnancy. She was married for 10 years and was under treatment for infertility since 7 years. The hysterosalpingogram aroused the suspicion of a bicornuate uterus though it was not confirmed laparoscopically. The present pregnancy was a natural conception. Ultrasonography done in the first trimester did not show anything abnormal. At 5 months into pregnancy, she had an episode of vaginal bleeding, when ultrasound revealed fetal demise. Termination of pregnancy using vaginal and systemic prostaglandins was unsuccessfully attempted by the practitioner, and patient referred to our centre for hysterotomy.

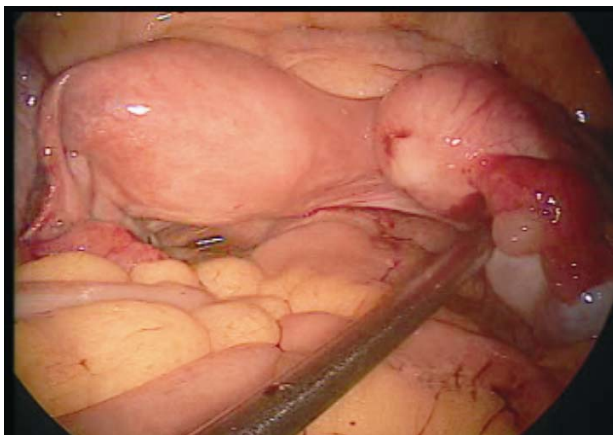


Figure 1. Laparoscopic view of rudimentary horn with normal uterus, causing CPP in Case 1. Laparoscopic hemi hysterectomy of the rudimentary horn was performed (**Figure 2**). The patient had an uneventful post operative period. Histopathological examination confirmed uterine tissue. The patient remains symptom free till date.



Figure 2. Altered blood which was aspirated from the horn and the resected rudimentary horn.

On *examination*, her vitals were stable; there was a palpable abdominal mass arising from pelvis corresponding to 14 wks gravid uterus. Speculum examination demonstrated a single centrally placed cervix. On vaginal examination, cervix uneffaced, uterus normal sized deviated to left side. A mobile nontender cystic mass 8 cm × 8 cm was felt on the right side. Ultrasound revealed a normal sized empty uterus having endometrial thickness of 5 mm. A gestational sac with a dead fetus of 15 wks was noted to the right and anterior to uterus (**Figure 3**).

Both ovaries were normal. Provisional diagnosis of secondary abdominal pregnancy was considered. MRI reported similar findings (**Figure 4**), further confirming the diagnosis of secondary abdominal pregnancy.



Figure 3. Ultrasound of Case 2 showing single fetus in a muscular structure outside the normal uterus.



Figure 4. MRI showing findings suggestive of abdominal pregnancy.

An exploratory laparotomy was performed. At laparotomy, the uterus, left tube and ovary were normal. There was a right sided 8 cm × 8 cm muscular structure, attached to uterus by a fibrous band; the right tube was seen arising from this mass. Round ligament of right side was seen arising lateral to this mass, thereby confirming it to be a uterine horn. The right ovary was normal. This rudimentary horn was excised with the fetus in situ. Cut section of the mass showed a muscular wall with a dead fetus inside. Post operative recovery of the patient was uneventful. Definitive confirmation of uterine tissue was

done by histopathology.

4. DISCUSSION

The exact incidence of mullerian abnormalities is not known because of normal fertility outcome in majority of these women. Estimates range from 1% - 3.5%, with higher percentage prevalence in infertile women (6.3%) [1]. Unicornuate uterus is categorized in Class II of the American Fertility Society classification (1988) of mullerian anomalies. It accounts for 2.4% - 13% of all mullerian anomalies [2,3]. Since fertility outcome is not usually affected, the clinical presentation may be highly variable; ranging from an apparently trivial dysmenorrhoea in an adolescent girl to intractable vague pelvic pain in a parous woman. The presence of normal menstrual flow further blankets the possibility of a uterine anomaly. As high as 78% of rudimentary horns have been known to present in the third decade of life or later, most often as acute emergencies resulting from rupture of horn pregnancy [4]. Unless specifically sought, ultrasound done for other gynecologic disorders often fails to pick up such malformations. In a review of 266 rudimentary horn presentations, sensitivity of two dimensional USG as a diagnostic tool was shown to be only 26% [4]. A novel method of prerupture diagnosis of rudimentary horn pregnancy has been suggested by Buntugu *et al.* The method involves placement of a Foley's catheter into uterine cavity with bulb inflated. An abdominal ultrasound with full bladder is then performed. Although the method is claimed to offer better sensitivity over conventional sonography, more studies are needed before it can be recommended for routine use [5]. Recent studies by Ghi *et al.*, found 3-dimensional transvaginal ultrasonography to be extremely accurate in the diagnosis of uterine anomalies. They found a concordance between endoscopy and 3D ultrasound in 52 of 54 cases [6]. However, MRI where available, still remains the gold standard for confirmation of mullerian anomalies.

Chronic pelvic pain has been reported in 15% - 20% of women aged 18 yrs - 50 yrs [7,8,9] and is an indication for at least 40% of all gynecologic laparoscopies [10]. Whereas 8.5% of adolescents undergoing laparoscopy for pelvic pain have some form of uterine malformation [11], mullerian anomalies as a cause for chronic pelvic pain in a parous woman is rarely reported [12].

We were intrigued by the late presentation and missed diagnosis of existence of this horn in our patient who had had a normal reproductive outcome including a cesarean section. The patient had denied any prior history of pelvic pain or dysmenorrhoea, suggesting recent activation of disease. Possible explanation for the late onset of symptoms in this case could be tubal ligation inciting

accumulation of menstrual blood in the horn, thereby precipitating pelvic pain. This phenomenon has also been reported by Fugimoto *et al* [13]. Rudimentary horn as a cause of pelvic mass and pain has even been reported after vaginal hysterectomy in a 41 yr old lady [14]. These reports suggest possible role of various "triggers" which cause activation and presentation of such rudimentary horns late in reproductive life. The diagnosis had been completely missed at cesarean section probably due to the small size of the horn as compared to the term gravid uterus and the obvious altered anatomic relations.

The commonly accepted treatment for this entity is excision through laparotomy or laparoscopy. Other treatment options include endometrial ablation of accessory horn through hysteroscopic approach; with reported symptom free period of up to 3 yrs [15]. Hysteroscopic drainage of hematometra in a noncommunicating accessory horn by using electrocautery to create a communication between the horns has also been described [16].

Pregnancy in rudimentary horn has been noted once in 1225 ectopic pregnancies [17] and once in 76000 total pregnancies [18]. The most significant threat is the risk of rupture; which is estimated to be about 50% and most commonly occurs in second trimester [19]. Various signs and symptoms pointing to presence of a uterine anomaly have been described, including past history of dysmenorrhoea, pregnancy along with a freely mobile tumor, passage of a decidual cast, absent tenderness on examination (unlike a tubal ectopic) [20] etc. But still, only a very small percentage of these rudimentary horn pregnancies are accurately diagnosed pre-operatively. While excision of this "ectopic" pregnancy along with the accessory horn remains the most common treatment modality, combined medical and surgical treatment has also been tried. This involves fetal intracardiac potassium chloride and intraplacental methotrexate, followed 6 wks later by laparoscopic resection of the horn [19]. Such combined medical, surgical modality has the advantage of reducing the vascularity, thereby decreasing operative blood loss [19]. Additional studies are needed before these treatment modalities are widely accepted.

5. RECOMMENDATIONS

The authors would like to recommend that clinicians be sensitive to the presence of mullerian anomalies in patients with otherwise normal gynecologic and reproductive histories. The diagnosis should be considered in unusual presentations of pelvic pain or early pregnancy. Although relatively insensitive, the routine ultrasound may prove helpful if clinicians bear in mind these differential diagnoses. Emergency staff should be alert to the possibility of such malformations in patients presenting with acute abdomen, suspected ruptured ectopic

or even failed induction with adequate doses of prostaglandins. A thorough inspection of the pelvis should be performed at the time of any operative procedure. Prophylactic resection of a noncommunicating uterine horn should be considered in patients with incidental discovery on laprotomy to prevent endometriosis and possible future horn pregnancy.

6. CONFLICT OF INTEREST

We declare that we have no conflict of interests.

REFERENCES

- [1] Simon, C., Martinez, L., Pardo, F., *et al.* (1991) Mullerian defects in women with normal reproductive outcome. *Fertility and Sterility*, **56**, 192-193.
- [2] Grimbizis, G.F., Camus, M., Tarlatzis, B.C., Bontis, J.N. and Devroey, P. (2001) Clinical implications of uterine malformations and hysteroscopic treatment results. *Human Reproduction*, **7**, 161-174.
- [3] Acién, P. (1997) Incidence of Müllerian defects in fertile and infertile women. *Human Reproduction*, **12**, 1372-1376.
- [4] Jayasinghe, Y., Rane, A., Stalewski, H. and Grover, S. (2005) The presentation and early diagnosis of the rudimentary uterine horn. *Obstetrics Gynecology*, **105**, 1456-1467. [doi:10.1097/01.AOG.0000161321.94364.56](https://doi.org/10.1097/01.AOG.0000161321.94364.56)
- [5] Buntugu, K.A., Ntummy, M.Y., Ameh, E.O. and Obed, S.A. (2008) Rudimentary horn pregnancy: Prerupture diagnosis and management. *Ghana Medical Journal*, **42**, 92-94.
- [6] Ghi, T., Casadio, P., Kuleva, M., Perrone, A.M., Savelli, L., Giunchi, S., *et al.* (2009) Accuracy of three-dimensional ultrasound in diagnosis and classification of congenital uterine anomalies. *Fertility and Sterility*, **92**, 808-813. [doi:10.1016/j.fertnstert.2008.05.086](https://doi.org/10.1016/j.fertnstert.2008.05.086)
- [7] ACOG Practice Bulletin #51 (2004) Chronic pelvic pain. *Obstetrics and Gynaecology*, **103**, 589-605.
- [8] Mathias, S.D., Kuppermann, M., Liberman, R.F., Lipschutz, R.C. and Steege, J.F. (1996) Chronic pelvic pain: Prevalence, health related quality of life, and economic considerations. *Obstetrics Gynecology*, **87**, 321-327. [doi:10.1016/0029-7844\(95\)00458-0](https://doi.org/10.1016/0029-7844(95)00458-0)
- [9] Jamieson, D.J. and Steege, J.F. (1996) The prevalence of dysmenorrhoea, dyspareunia, pelvic pain, and irritable bowel syndrome in primary care practitioners. *Obstetrics Gynecology*, **87**, 55-58. [doi:10.1016/0029-7844\(95\)00360-6](https://doi.org/10.1016/0029-7844(95)00360-6)
- [10] Howard, F.M. (1993) The role of laparoscopy in chronic pelvic pain: Promise and pitfalls. *Obstetrical and Gynecological Survey*, **48**, 357-387. [doi:10.1097/00006254-199306000-00001](https://doi.org/10.1097/00006254-199306000-00001)
- [11] Vercellini, P., Fedele, L., Arcaini, L., *et al.* (1989) Laparoscopy in the diagnosis of chronic pelvic pain in adolescent women. *Journal of Reproductive Medicine*, **34**, 827.
- [12] Howard, F.M. (2003) The role of laparoscopy in chronic pelvic pain patient. *Clinical Obstetrics and Gynecology*, **46**, 749-766. [doi:10.1097/00003081-200312000-00004](https://doi.org/10.1097/00003081-200312000-00004)
- [13] Fugimoto, V.Y., Klein, N.A. and Miller, P.B. (1998) Late onset hematometra and hematosalpinx in a woman with a noncommunicating uterine horn: A case report. *Journal of Reproductive Medicine*, **43**, 465-467.
- [14] Stitely, M.L. and Hopkins, K. (2006) Laparoscopic removal of a rudimentary horn in a previously hysterectomized patient. *Journal of the Society of Laparoendoscopic Surgeons*, **10**, 257-258.
- [15] Hucke, J., DeBruyne, F., Campo, R.L. and Freikha, A.A. (1992) Hysteroscopic treatment of congenital uterine malformations causing hemihematometra: A report of three cases. *Fertility and Sterility*, **58**, 823-825.
- [16] Nogueira, A.A., Candido dos Reis, F.J. and Campolungo, A. (1999) Hysteroscopic treatment of unicornuate uterus associated with a cavitary rudimentary horn. *International Journal of Gynecology and Obstetrics*, **64**, 77-78. [doi:10.1016/S0020-7292\(98\)00160-X](https://doi.org/10.1016/S0020-7292(98)00160-X)
- [17] Jarcho, J. (1946) Malformations of the uterus; review of subject, including embryology, comparative anatomy, diagnosis and report of cases. *American Journal of Surgery*, **71**, 1946.
- [18] Kadan, Y. and Romano, S. (2008) Rudimentary horn pregnancy diagnosed by ultrasound and treated by laparoscopy a case report and review of literature. *Journal of Minimally Invasive Gynecology*, **15**, 527-530. [doi:10.1016/j.jmig.2008.05.010](https://doi.org/10.1016/j.jmig.2008.05.010)
- [19] Park, J.K. and Dominguez, C.E. (2007) Combined medical and surgical management of rudimentary uterine horn pregnancy. *Journal of the Society of Laparoendoscopic Surgeons*, **11**, 119-122.
- [20] Daskalakis, G., Athanasios, P., Katerina, L. and Aris, A. (2002) Rupture of noncommunicating rudimentary uterine horn pregnancy. *Obstetrics and Gynecology*, **100**, 1108-1110. [doi:10.1016/S0029-7844\(02\)02153-1](https://doi.org/10.1016/S0029-7844(02)02153-1)