A study of complications in case of unicornuate uterus with rudimentary horn

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ABSTRACT

Background: To analyse gynecological and reproductive morbidities associated with unicornuate uterus with noncommunicating rudimentary horn.

Methods: This is a retrospective study of 20 cases of unicornuate uterus with noncommunicating rudimentary horn found on laparotomy in a duration of 5 years (Oct 2011-Oct 2016).

Results: Out of 20 patients, two teenagers presented with dysmenorrhoea and pain abdomen and had haematometra in the noncommunicating rudimentary horn which was excised. Eight had pregnancy in the noncommunicating rudimentary horn of which all presented after rupture and five were admitted in a state of shock. Ten patients had pregnancy in the hemiuterus with complications inherent to the condition.

Conclusions: Unicornuate uterus with noncommunicating rudimentary horn is associated with poorest outcomes among all uterine anomalies and a high index of suspicion is needed to diagnose this condition and thus to save the woman from catastrophic complications.

Keywords: Mullerian, Rudimentary, Rupture, Unicornuate

INTRODUCTION

Uterine malformations are the results of abnormal mullerian duct development-fusion, canalization and septal defects. Unicornuate uterus with a rudimentary horn is one such anomaly of uterus occurring due to fusion defects. It belongs to class-II mullerian anomalies according to classification by American fertility society.1

This malformation is rare and it 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 of the menstrual flow may cause hematomata, leading to endometriosis and infertility. 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.2,3

Conception in the rudimentary horn is very rare, and occurs either from small communication with uterine cavity or by trans-peritoneal migration of the sperm from contralateral side.4 Diagnosis may be suspected by clinical examination-1) Bimanual palpation of a mass extending outside the uterine angle (Baart’s de la Faille’s sign); 2) displacement of the fundus to the contralateral side with rotation of the uterus and elevation of the affected horn (Ruge Simone syndrome); and 3) deviation of uterus to one side with an adnexal mass in pregnancy may indicate the presence of rudimentary horn.5 Hysterosalpingography, ultrasonography and magnetic
resonance imaging may be useful in diagnosis of unicornuate uterus. HSG shows a deviated ‘banana-shaped’ uterus with single fallopian tube. Trans-vaginal sonography may show a small, well-formed elliptical uterus with single cornu deviated to one side. 3-D imaging and MRI may give a classic banana picture. Renal anomalies are detected in 40% of cases. Variable thickness of rudimentary horn musculature, dysfunctional endometrium and poor distensibility of the myometrium lead to rupture of the rudimentary horn. 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. 6,7

With this background, a study was conducted where all cases of unicornuate uterus with rudimentary horn found on laparotomy were analysed to know their gynaecological and obstetric implications.

METHODS

This is a study of 20 cases diagnosed at laparotomy at a tertiary care teaching hospital over a period of 5 years from Oct 2011-Oct 2016. which were retrospectively analyzed. These patients were divided into 2 groups: group-I (n=2) consisted of patients who presented with gynaecological complaints and in whom the condition was diagnosed at laparotomy. Group-2 (n=18) consisted of gravid patients, Group-2a (n=8) consisted of pregnancy in rudimentary horn of uterus group-2b (n=10) consisted of patients who had pregnancy in the semi uterus (normal horn) and the rudimentary horn happened to be an incidental finding during LSCS for some obstetric indication.

RESULTS

Group 1-the age of the two patients in this group was 18 and 13 years respectively. The first patients were referred with complain of severe pain abdomen from a peripheral hospital and USG done there showed uterine anomaly! - double uterus. CT Scan done at our centre conformed the diagnosis and showed blood clots in one horn of uterus. Laparotomy was done which showed the rudimentary horn with haematometra. It was a non-communicating horn with a fibromuscular band of fusion with the other half of uterus. Resection of the rudimentary horn was done.

Another case was that of a 13 years old girl who had attained menarche 2 years back and had undergone Appendicectomy 1 year back at a peripheral hospital but the pain of right iliac fossa (for which appendicectomy was done) worsened. USG also did not point towards the unexpected cause of lingering pain. Diagnostic laparoscopy gave the diagnosis of rudimentary noncommunicating horn of uterus which was resected and was found to be having haematometra of about 75 ml. In both cases the non-communicating rudimentary horn was of the right side.

Table 1: Pregnancy outcome in rudimentary horn pregnancy and its characteristics.

<table>
<thead>
<tr>
<th>Age (year)</th>
<th>Obstetric history</th>
<th>Duration of gestation</th>
<th>Clinical Diagnosis</th>
<th>Preoperative Diagnosis (USG+Clinical)</th>
<th>Location of rudimentary horn</th>
</tr>
</thead>
<tbody>
<tr>
<td>27</td>
<td>G2P1 L1</td>
<td>12</td>
<td>Pain abdomen USG showed ectopic pregnancy with haemo-peritonium</td>
<td>Ectopic pregnancy</td>
<td>Right side</td>
</tr>
<tr>
<td>23</td>
<td>Primi</td>
<td>26</td>
<td>Pain in Abdomen, Shock</td>
<td>Rudimentary horn pregnancy</td>
<td>Right side</td>
</tr>
<tr>
<td>28</td>
<td>G2P1 L1</td>
<td>27</td>
<td>Vomiting, diarrhea, pain abdomen shock, history of fall</td>
<td>? Rupture uterus? Abruption</td>
<td>Right side</td>
</tr>
<tr>
<td>24</td>
<td>Primi</td>
<td>24</td>
<td>Pain abdomen, shock</td>
<td>Rupture of Rudimentary horn pregnancy</td>
<td>Right</td>
</tr>
<tr>
<td>22</td>
<td>G3P1 L1 A1</td>
<td>16</td>
<td>Pain abdomen, secondary infertility</td>
<td>Rupture of cornual pregnancy</td>
<td>Left</td>
</tr>
<tr>
<td>32</td>
<td>G3P2L2 with previous LSCS</td>
<td>17</td>
<td>Pain abdomen, shock</td>
<td>Rupture uterus/scar pregnancy</td>
<td>Left</td>
</tr>
<tr>
<td>30</td>
<td>G2A1</td>
<td>22</td>
<td>Vaginal bleeding, pain abdomen</td>
<td>Rudimentary horn pregnancy</td>
<td>Right</td>
</tr>
<tr>
<td>29</td>
<td>G2P1 L1</td>
<td>28</td>
<td>Pain abdomen, shock</td>
<td>Rudimentary horn pregnancy</td>
<td>Right</td>
</tr>
</tbody>
</table>
**Case 2 a**

8 patients had pregnancy in the non-communicating rudimentary horn, the data of these patients is given in table 1. One patient presented in the first trimester and 7 patients in the 2nd trimester. Pain and shock were the main presenting complain with varying degree of pallor. Preoperative diagnosis of rudimentary horn pregnancy was suspected in two cases only and in the remaining, the diagnosis was made at laparotomy. The malformation was on the right side in 6 cases and left side in two. All women underwent excision of the rudimentary horn with ipsilateral salpingoopherectomy.

**Case 2 b**

We witnessed non-communication rudimentary horn in 10 cases as incidentaloma during LSCS which was mostly done formal presentation and in coordinate uterine actions.

In 4 of the above cases-renal anomalies in the form of pelvic kidney and aberrant fused renal mass was seen in USG.

**Table 2: Pregnancy outcome in unicornuate uterus (hemiuterus).**

<table>
<thead>
<tr>
<th>Pregnancy outcome</th>
<th>No. of pregnancies n=10</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Abortion (previous pregnancy)</td>
<td>2</td>
<td>20</td>
</tr>
<tr>
<td>Cerclage required</td>
<td>3</td>
<td>30</td>
</tr>
<tr>
<td>Preterm</td>
<td>2</td>
<td>20</td>
</tr>
<tr>
<td>Term</td>
<td>5</td>
<td>50</td>
</tr>
<tr>
<td>Breech</td>
<td>4</td>
<td>40</td>
</tr>
<tr>
<td>Transverse lie</td>
<td>1</td>
<td>10</td>
</tr>
<tr>
<td>IUGR</td>
<td>2</td>
<td>20</td>
</tr>
<tr>
<td>Oligohydramnios</td>
<td>3</td>
<td>30</td>
</tr>
<tr>
<td>Incoordinate Uterine action</td>
<td>2</td>
<td>20</td>
</tr>
<tr>
<td>IUD</td>
<td>1</td>
<td>10</td>
</tr>
<tr>
<td>Live births</td>
<td>8</td>
<td>80</td>
</tr>
<tr>
<td>Early neonatal death</td>
<td>1</td>
<td>10</td>
</tr>
</tbody>
</table>

**DISCUSSION**

It is difficult to truly estimate the incidence of these complications as the data available are in the form of case reports and surveys collected from the literature that usually has only the severe cases requiring surgery. High index of suspicion should be kept in teenagers presenting with dysmenorrhea. To decrease the serious complications in future, early diagnosis is of utmost importance.

The patient should be treated by excision 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. In our analysis, hematometra was seen in two cases only.

Five of our patients presented in a state of shock due to rudimentary horn rupture. The duration of the pregnancy at the occurrence of rupture are dependent on the thickness of the myometrium. If the pregnancy grows, it usually overcomes the first trimester period uneventfully as the rudimentary horn is thicker than the fallopian tube and 80-90% of the ruptures in the second trimester. As it is associated with such catastrophe every effort should be made to diagnose it in early pregnancy but according to the literature, less than 5% of the reported cases were diagnosed preoperatively and mostly the diagnosis was made at laparotomy or laparoscopy.

A careful pelvic examination in the 1st trimester showing deviated uterus with palpable contra-lateral pelvic adnexa should arouse suspicion of uterine anomaly. Ultrasoundography can also pick up this anomaly with reasonable accuracy. A gestational sac surrounded by myometrium by the side of a normal empty uterus and non-communication of gestational sac with the endometrial cavity and the cervix differentiates it from pregnancy in one of the horns of a bicornuate uterus. MRI and CT scan are also gaining popularity for diagnosing uterine malformations. Both clinically and radiologically the diagnosis is more accurate in the early first trimester when the two horns are separate in the pelvis.

Once the diagnosis is strongly suspected these patients should be taken up for laparoscopy or laparotomy, depending upon the general condition of the patient, and the rudimentary horn should be excised along with its tube, so as to prevent future tubal ectopic pregnancy in that tube.

In present study, preoperative diagnosis was possible on clinical and sonography findings in only four out of the eight patients, and that too because of high index of suspicion as in our hospital the relative occurrence of complications arising out of rudimentary horn is higher than its occurrence in the general population for unexplained reason. In a study by Goel et al pre-operative diagnosis was suspected in 2/7 cases. In our study one case was seen in 1st trimester as ectopic pregnancy, and seven cases were seen in 2nd trimester. The rudimentary horn should be excised whenever diagnosed. Laparotomy with resection of rudimentary horn was done in all rudimentary horn pregnancies in present study similar to study by Vani Malhotra et al. Indications for resection (Anne deviwold, 2006) are rupture, unilateral dysmenorrhea, and haematometra. In all such cases, evaluation of renal system is indicated because of the high incidence of associated urological anomalies.

In present study, ten pregnancies occurred in unicornuate uterus with rudimentary horn and diagnosis was made
incidentally at LSCS similar to studies by Vani Malhotra and Goel et al.\textsuperscript{12,11} All obstetric complications like abortion, preterm delivery, IUGR, IUD, were more compared to normal, similar to study by Goel et al.\textsuperscript{11} Rate of abortion was 20%, similar to 25.8% in Goel et al and 24.3% in Reichman et al.\textsuperscript{11,14} Rate of pre-term delivery was 20% similar to 20.1% in Reichman et al.\textsuperscript{14} 10% had intrauterine fetal demise compared to 3.8% in Reichman et al.\textsuperscript{14} Pregnancy outcome in present study was poor overall. However, rate of term delivery was 50%, compared to 32.25% in Goel et al may be because of good antenatal care overall. Live birth rate was 80%.\textsuperscript{11}

**CONCLUSION**

Unicornuate uterus with noncommunicating rudimentary horn is associated with poorest outcomes among all uterine anomalies and a high index of suspicion is needed to diagnose this condition and thus to save the woman from catastrophic complications.

**ACKNOWLEDGMENTS**

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Ethical approval: The study was approved by the Institutional Ethics Committee

**REFERENCES**