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A Silent Rupture of Extra Horn of Bicornuate Uterus, Due to Molar Pregnancy

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Abstract: ***Molar pregnancy is an abnormality in chorionic villi that cause trophoblastic proliferation and edema of villous stroma. Ectopic molar preganancy is a very rare entity. Unicornuate uterus with rudimentary horn occurs due to failure of complete development of one of the mullerian ducts and incomplete fusion with contralateral side. Pregnancy in non communicating rudimentary horn is extremely rare and usually terminates in rupture during first and second trimester of pregnancy. Variable thickness of rudimentary horn musculature, poor distensibility of myometrium leads to rupture. Diagnosis of rudimentary horn is difficult and can be missed in ultrasonography. This report literature is replite with a unique case report of ruptured horn of bicornuate uterus in case of complete molar pregnancy, a rare and unreported occurrence so far.***

Keywords: Bicornuate Uterus, Molar Pregnancy

1. Introduction

Molar pregnancy is an abnormality in chorionic villi that causes trophoblastic proliferation and edema of villous stroma. Molar pregnancy is common in Asian countries, incidence is about 1 in 100 pregnancies, in U.S it is 1 in 1000 pregnancies[1]. Ectopic pregnancy incidence is about 20 per 1000 pregancies[2], but ectopic molar pregnancy is very rare. The uterine horn is the least frequent site for location of ectopic pregnancies, with 2.4% of cases[3].

Bicornuate uterus (Bicornis-Unicollis) is a developmental anomaly, due to failure of fusion in upper part of paramesonephric duct, presents as a double uterus with a single cervix and vagina. It occurs in 0.1 -0.5% of women[4]. Of all mullerian agenesis, incidence of bicornuate uterus is 25%.

Incidence of pregnancy in rudimentary horn is 1 in 40, 000 to 1, 50, 000 pregnancies[5].

Incidence of uterine rupture in pregnancy is 0.07%[6] and Spontaneous rupture of unscarred uterus is 0.012%[7].

A case of uterine rupture due to Molar pregnancy is hitherto unknown, & rupture of an extra horn of a bicornuate uterus, has not surfaced so far in literature of reported cases.

2. Case Report

The literature is replite with a unique case report of ruptured horn of bicornuate uterus in case of complete molar pregnancy, a rare and unreported occurrence so far.

A patient, 22yr old female, primigravida, on 31-05-17, was referred with USG Abdomen- pelvis report, revealing Complete molar pregnancy and USG Chest in sitting position, revealing mild free fluid in left side of pleural cavity, to emergency department of Dhiraj Hospital, Vadodara, with amenorrhoea of 6moths and pain in abdomen since 5 days.

She was married 9 months ago. Her menstrual cycle was regular. Last menstrual period was not known. She had irregular antenatal check-ups at primary health care.

The patient was conscious, cooperative and oriented to time, place, person. Patient was afebrile, pale and icteric with pulse 104/min, BP-120/78 mmHg.

Obstetric examination findings were as follows-

Per abdominal examination revealed uterus of 20 wk size and tenderness in left iliac fossa without guarding and rigidity. Per vaginal examination elicited tenderness in both fornices, more in left, than on right side.

USG Abdomen Pelvis revealed uterus with molar pregnancy & rupture of uterus on anterior wall of fundus with extension of hydratidiform mole in pelvic cavity with moderate fluid in pelvic cavity.

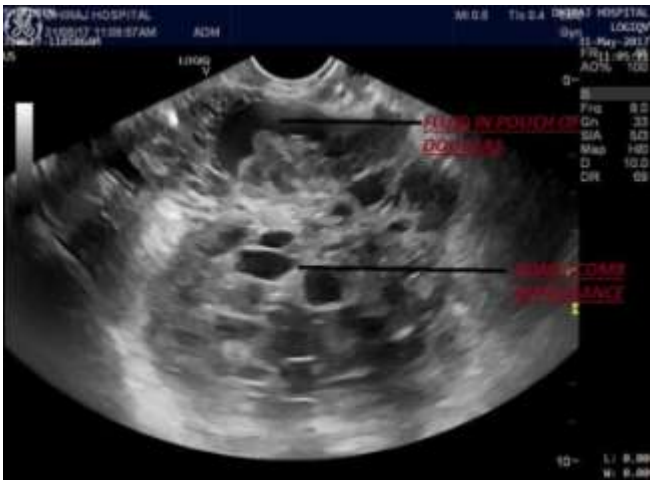

Figure 1: USG shows honey comb appearance

Urine Pregnancy Test was positive. Further routine investigations were done, her haemoglobin concentration was 7.8gm% and Beta hCG was 50, 000 u/l and TSH was1.8mIu/l. Reports of other investigations were normal.

Patient was shifted to operation theatre for Laparotomy under general anaesthesia. Median incision was made extending from 2cm below the umbilicus up to pubic symphysis, of about 10cm length. Subcutaneous fat and rectus sheath were opened by sharp and blunt dissection, parietal peritoneum was opened.

Intraop Findings- Haemo-peritoneum was present, about 300 ml blood in cavity. Small grape of, size5mm to 30mm, like vesicles were present in clusters. Uterus was bicornuate with one well developed horn, and a small horn, both with single cervix.

Grape like vesicles coming out from posterior surface of fundus of left horn. Left side horn was dissected and left side fallopian tube was reconstructed for maintaining fertility. Left side horn was non- communicating, it acts like a rudimentary horn. Right side fallopian tube was not seen. Both side ovaries were normal. Grape like structure were adherent to Omentum. The specimen sent for Histology.

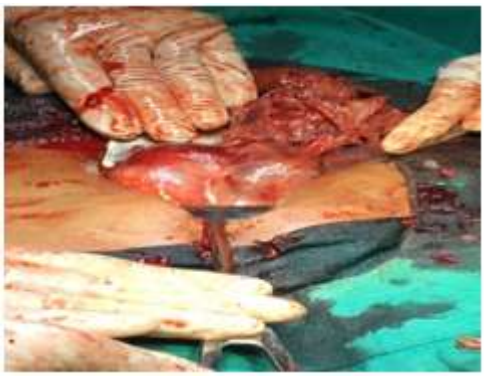

Figure 2: Bicornuate uterus with vesicular mole

4 units of packed cell volume and 2 units free frozen plasma were transfused. Post operatively patient was given iv fluids, antibiotics, analgesics and antacids. Beta HCG was repeated on 3rd postoperative day, its value had reduced to 6000u/l, repeated on 8th postoperative day, it reduced up to 3056u/l.

Stitches were rermoved on 8th postoperative day and patient was discharged. As patient residing at a far place, she did not come for follow up. Hence, follow up with repeated Beta-HCG esimation could not be carried out. Patient was lost for a follow up, as she did not respond to our telephone calls, as well. As a result, further management with chemotherapy, could not be envisaged. We can simply hope for best, that it all will end well, for the patient, in the long run.

She was counselled about her uterine anomaly. She was advised to avoid pregnancy for 2 years, using barrier contraceptives.

She was advised to come for antenatal check up as soon as she as concieves in future.

Histology report-Histologically, Edematous villi surrounded circumferrentially by Marked proliferation of the outer syncytio-trophoblast and cytotrophoblastic epithelium. Trophoblast show round to oval uniform nuclei with no nuclear atypia and abundant eosinophillic cytoplasm. Histologically, no evidence of malignancy.

3. Discussion

Ectopic molar pregnancy is extremely rare, and preoperative diagnosis is difficult. More-ever, the efficacy of trans-vaginal sonography in diagnosis of ectopic molar pregnancy is very poor. It confirmation is possible during operation or by histopathology study. We carried out a review of all ectopic molar pregnancy cases, published in English from 1996-2017. All studies were obtained from Medline using the term “ectopic molar pregnancy”, and from references of the articles. We identified 26 articles, reporting 31 cases, of ectopic molar pregnancy[8]. All literature demonstrate, ectopic molar pregnancy can be found in the fallopian tube (61%), ovary (16%), cornu (10%), peritoneum (6%), uterine cervix (3%), and caeserian scar (3%). Twenty one cases presented with rupture and haemoperitoneum. The rate of rupture of ectopic molar pregnancy is 67%.

Rudimentary horn pregnancy occurs 1 in 76, 000. In 70-90% of cases, the horn is non- communicating[9]. Mostly it is diagnosed during surgery, when a mishap occurs.

The first rudimentary horn pregnancy was described by Vassal and Mauriceau in 1669[10]. Rudimentary horn pregnancy results in rupture of horn in 80-90% of cases by 2nd trimester[11], and only in about10% cases, a full term pregnancy results. Rudimentary horn pregnancy represents a serious obstetric condition & it’s rupture poses as a life threatening obstetric complication.

Rupture of horn in bicornuate uterus in molar pregnancy occurs because of inability of malformed uterus, to expand as a normal uterus. Uterine rupture may occur due to weak and deficient musculature of the anomalous uterus, or it could be due to narrow communication between left horn and cervix, molar pregnancy due to higher invasion of trophoblastic proliferation, could not accommodate in a small horn, led to rupture of uterus. Rupture of the horn does not usually take place until the twelfth week of gestation but, when it does, there is nearly always severe bleeding which makes the condition extremely dangerous. The present case is of a silent rupture, the site of rupture in present case is the fundus of the uterus, which is also very rare.

Other causes of fundal rupture are due to injury, during previous dilatation and curettage, placenta percreta and fundal pressure during 2nd stage of labour.

4. Conflict of Interest

There is no conflict of interest

5. Foot Note

This case report is presented here, just because of rarity of such an obstetric calamity, occurring hardly once in the practicing life of an obstetrician.

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