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## Case Report

# Ectopic molar pregnancy: A rare entity

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### ABSTRACT

Ectopic molar pregnancy is a relatively rare condition and is reported in about 1.5 every 1,000,000 pregnancies. Ectopic molar pregnancy mimics tubal ectopic pregnancy in clinical features, and treated surgically. We report a case of a woman who was diagnosed with an unruptured tubal pregnancy and underwent laparotomy proceed bilateral salpingectomy. Her serum Bhcg prior to surgery was 74768 IU/ml. Grossly the ectopic pregnancy tissue was that of a molar pregnancy, which was confirmed by histopathology as partial molar pregnancy. She was followed up with serial serum Bhcg till normal. Tubal molar pregnancy is a rare entity and a clinicopathological diagnosis.

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## 1. Introduction

Ectopic pregnancy is an abnormal implantation of fertilized ovum outside the uterine cavity occurring in 20 per 1,000 pregnancies.<sup>1</sup> Molar pregnancy occurs due to improper fetal and placental development with an incidence of 1 in 500 to 1000 pregnancies.<sup>2</sup> While ectopic pregnancy and molar pregnancy are not rare, an ectopic molar pregnancy is an extremely rare event. There have only been a few cases of molar ectopic pregnancies reported in the literature with an incidence of 1.5 in every 1,000,000 pregnancies.<sup>3</sup> Ectopic molar pregnancy is an extremely rare entity, which can occur at any place in the pelvic cavity. The first report on tubal mole was made by Otto in 1871.<sup>4</sup> A literature review of 31 cases of ectopic molar pregnancy demonstrated that lesions have been found in the fallopian tube (19 cases, 61%), ovary (5 cases, 16%), cornu (3 cases, 10%), peritoneum (2 cases, 6%), uterine cervix (1 case, 3%), and cesarean scar (1 case, 3%).<sup>5</sup>

In this, we present a case report of unruptured tubal molar pregnancy, who reported to our hospital was

hemodynamically stable, after clinical examination and USG was confirmed of unruptured right ectopic she underwent surgery. Unruptured right ectopic was sent for histopathological examination which confirmed partial molar pregnancy. Hence, her follow up was done with serial beta hcg as in case of molar pregnancy until a negative betahcg was obtained. Thus, the need for histopathology follow up is a necessity for diagnosis, treatment and follow up of such tubal molar pregnancy. Need for correct diagnosis is more important as the complications, treatment modalities and follow up varies.

## 2. Case Report

A 35-year old female patient with two living children born out of previous two Lower segment Caesarean sections presented to our department with complaints gradually worsening abdominal pain since past 15 day, with two episodes of vomiting since past two days and bleeding per vagina since 9 days, which was preceded by three months of amenorrhoea. Her Urine pregnancy test was positive at 98<sup>th</sup> day of missing period. She gave no history of any abnormal discharge per vaginum or passage of grape like vesicles. She

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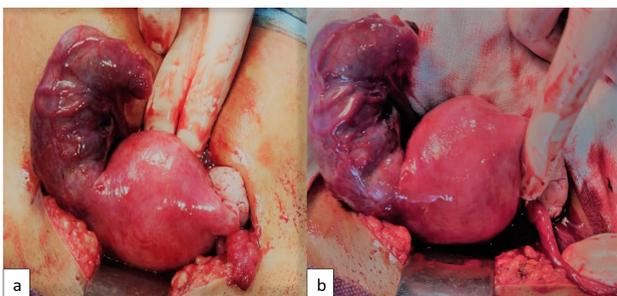
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gave no history of fainting episodes ten years ago along with her previous Caesarean section. There was no significant personal or family history.

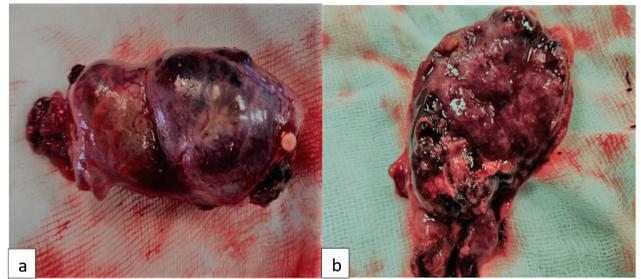
With the complaint of bleeding per vagina, she had consulted a local practitioner who performed an ultrasound and diagnosed her to have right unruptured ectopic pregnancy and advised her to visit tertiary care centre. On arrival to our emergency services, she was hemodynamically stable. Her pulse rate was 88 bpm and blood pressure was 110/80 mmHg. She had no pallor or lymphadenopathy. Abdominal examination revealed pfnestel scar and severe tenderness present in right iliac fossa. There was minimal bleeding per vagina noted, Uterus bulky anteverted, Cervix and vagina were healthy right sided fornicial fullness and tenderness present, left side fornix normal and non tender. A provisional diagnosis of unruptured right tubal pregnancy or tubal abortion was made on USG findings performed soon after admission. USG(TVS) revealed Right sided mass (6.08 x 4.21cm). No vascularity. Unruptured right tubal pregnancy or tubal abortion. Bulky uterus. POD with moderate free fluid. Beta -hcg was sent on admission revealed 74768 IU/ml. CBC Hb:10.8 g/dl, TC: 6500, S. Creat : 0.74mg/dl.

The patient was planned for exploratory laparotomy where Right sided unruptured tubal pregnancy of 8 x 3cm size in fibria was identified and fallope rings on both tubes indicated a previous laparoscopic tubectomy.(Figure 1) Bilateral salpingectomy was done in view of completed family and risk reduction of future ectopic pregnancy. Samples were sent to histopathological analysis.

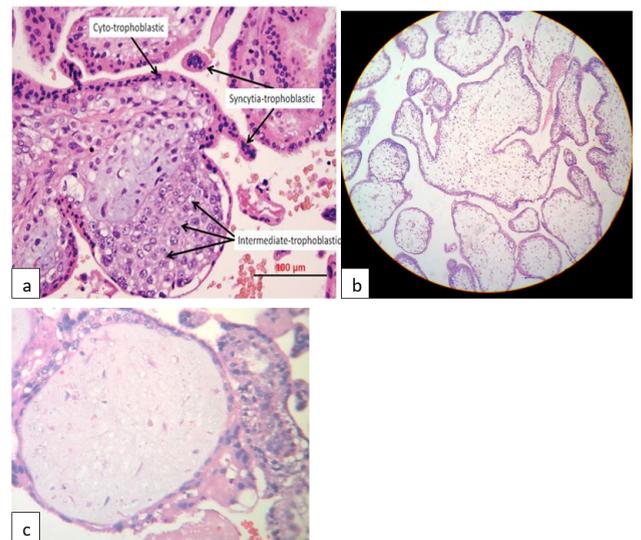
Cut section of the ectopic pregnancy showed trophoblastic tissue with no obvious gestational sac (Figure 2). Histopathological examination Variable sized villi with trophoblastic cells, Scalloping of chorionic villi and a positive staining in IHC which was suggestive of a Partial molar pregnancy.<sup>6,7</sup> (Figure 3) Her intraoperative and postoperative period was uneventful and she was discharged well on post operative day 4. The patient was followed up with weekly quantitative  $\beta$ -hCG titers until three consecutive  $\beta$ -hCG levels were negative.



**Figure 1:** a): Intraoperative finding in exploratory laparotomy: Right sided unruptured tubal pregnancy; b): Left sided normal tube



**Figure 2:** Cut section: plenty of trophoblastic tissue seen. No obvious gestational sac seen



**Figure 3:** Histopathological examination suggestive of partial molar pregnancy a): Variable sized villi with trophoblastic cells, Scalloping of chorionic villi; b) and c): Shows focal edema Degenerated villi with increased trophoblastic cells

### 3. Discussion

Molar pregnancy is an abnormal gestation characterized by the presence of hydropic change in the placental villi, with circumferential proliferation of trophoblasts. Nonmolar hydropic abortions are common; it is important clinically to distinguish molar pregnancies from nonmolar hydropic changes, because the former has suspicion of persistent trophoblastic disease.<sup>3,8</sup> Additionally, the blighted ovum is a common feature seen in ectopic pregnancy and can be misinterpreted as a true hydatidiform mole.<sup>9</sup> However, it is possible to identify the early swellings of the placental villi don't form a true hydatidiform mole.

The salient diagnostic criteria of nonmolar pregnancy are liquefaction and edema of villous stroma, scantiness or absence of the villous blood vessels, and trophoblastic proliferation.<sup>10</sup>

Diagnosis of moles histopathologically only by evaluation of hematoxylin and eosin stained slides is challenging, with poor interobserver reproducibility.<sup>11</sup> However can be added on with p57 IHC to distinguish between complete and partial mole and other ancillary methods.

Newer studies shows DNA flow cytometry, STR genotyping can be used as a complement for diagnosis by determining the ploidy, but with the limiting factor of lack of availability of these ancillary techniques at all centers.<sup>11,12</sup>

Histopathological features of molar pregnancy with partial moles has less edema and trophoblast proliferation. The presence of fetal tissue, with nucleated red blood cells or fetal membranes, suggests a partial mole over a complete mole, but rare complete moles have evidence of fetal tissue, including a complete mole with a twin, or a mosaic complete mole.<sup>6</sup> In partial moles, villi are more irregular and scalloped, with trophoblast inclusions which differs from the more rounded villi of complete moles.<sup>7</sup>

Ectopic molar pregnancy is a rare entity, and the diagnosis may often be overlooked as it mimics tubal ectopic pregnancy. Abdominal pain and abnormal vaginal bleeding were reported in 70% and 61% of the patients, respectively in tubal molar pregnancy. Twenty-one cases (67%) presented with rupture and hemoperitoneum.<sup>5</sup> The rate of rupture and hemoperitoneum in cases of molar ectopic pregnancy rupture was reported as 67%. Berlingieri et al. and Frates et al. reported rates of ruptured normal ectopic pregnancy of 29.5% and 25.2%, respectively.<sup>13,14</sup>

Ultrasonography of GTD is a heterogeneous, hypochoic, solid mass with cystic vascular spaces.<sup>15</sup> Ultrasonography has limited role in identifying the molar pregnancy as there is no ultrasound criteria for distinguishing ectopic tubal pregnancy from tubal molar pregnancy.

Beta-hcg levels can not be completely be used to distinguish due to lower values than in intrauterine molar pregnancy as there is inadequate vascularisation in tubal pregnancy. And even in a clinical case and results of literature search done in comparison in the last ten years on tubal mole pregnancy, the increase of  $\beta$ hCG was found to be similar between tubal ectopic pregnancies and tubal molar pregnancies, hence could not be considered a valid tool for the diagnosis.<sup>16</sup> Though clinical diagnosis of a tubal molar pregnancy is difficult, correlation with serum B-Hcg may raise a suspicion as it is usually >100000 IU/ml in molar pregnancies.

Histopathological diagnosis is the gold standard and is essential in order to follow up the patient and thereby prevent a persistent trophoblastic disease or diagnose it early during follow up. Hence making the preoperative diagnosis of tubal molar pregnancy difficult to distinguish from tubal ectopic pregnancy.<sup>17</sup>

After confirmation of the diagnosis, monitoring the patient to rule out any development of GTN, to detect it only

by signs and symptoms of metastatic disease; however, the likelihood of this possibility is uncommon.<sup>18</sup> Explorative laparoscopy or laprotomy considering the stability of the patient, with or without salpingectomy, is considered the gold standard of treatment in ectopic pregnancies.<sup>19</sup> When salpingectomy is performed, a histological diagnosis should be followed up so that the follow up treatment could be planned.

Follow up with serial BHCG is crucial as in any other molar pregnancy.

#### 4. Conclusion

Chronic Ectopic Molar pregnancy relatively a rare entity and is a Clinico pathological diagnosis. UPT for any patient with amenorrhoea irrespective of tubectomy status in reproductive age is always advised. Serum Beta-hcg levels to determine the next line of management especially in unruptured ectopic pregnancy and correlate the levels to predict molar pregnancy. TVS irrespective of Beta-hcg levels, to correlate clinically. Any Suspicion of molar pregnancy salpingectomy must be preferred. Histopathological examination stands out to be the gold standard for diagnosis with difficulty in differentiating ectopic tubal pregnancy from tubal molar pregnancy as there are no clinical, laboratory, or ultrasound criteria.

Lastly, though a rare entity, ectopic molar pregnancy needs adequate follow up after treatment, with serial B-Hcg just as in a case of uterine molar pregnancy.

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#### 6. Conflict of Interest

Nil.

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