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Case Report

A Rare Case Of Extra Uterine Complete Molar Pregnancy Transforming To Choriocarcinoma: A Case Report

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ABSTRACT

Gestational trophoblastic diseases include complete and partial molar pregnancy, invasive mole, placental site trophoblastic tumor, and choriocarcinoma. The most malignant form is Gestational choriocarcinoma. The present case is an extremely rare case of choriocarcinoma developed after primary complete mole present at the fimbrial end of fallopian tube. Clinical diagnosis was made as ruptured ectopic. Exploratory laprotomy showed a large ulcerative lesion covered with clot present on the posterior aspect of serosal surface of uterus along with numerous grapes like vesicles. On histopathological examination the final diagnosis of extra uterine complete mole transformed into choriocarcinoma was made.

KEYWORDS: Ectopic pregnancy, Gestational trophoblastic disease, Complete molar pregnancy, Choriocarcinoma, Beta HCG.

INTRODUCTION

The World Health Organization classifies gestational choriocarcinoma as the malignant tumor of gestational trophoblastic tissue. It is a rare form of cancer which occurs in the female genital tract with a very high propensity to metastasize to various sites, including lungs, vagina, brain, liver, kidney, and gastrointestinal tract, (in descending order of frequency)[1]. Approximately 30 % cases of Choriocarcinoma have metastatic disease at the time of diagnosis [2]. It usually associated with molar pregnancy, ectopic pregnancy, or abortion but may develop after normal pregnancy [3]. It is known to occur in 1 in 5333 tubal pregnancies and 1 in 1.6 million normal intrauterine pregnancies [4]. Choriocarcinoma developing in ectopic locations are usually more aggressive and associated with distant metastasis [5]. In most cases choriocarcinoma is associated with a positive pregnancy test even without pregnancy and with high levels of the beta human chorionic gonadotrophin hormone (β-hCG).

CASE REPORT

A 37 year old female, gravid 3 Para 1 0 1 1 was admitted in the emergency ward of Dr RPGMC, Tanda in condition of shock, history of pain abdomen for 1 day and bleeding per vaginum for 10 days. According to the woman's medical records, 3 month back she had one month amenorrhea, for which she visited private hospital. Urine test for pregnancy was positive. Patient consumed some pills to terminate the pregnancy. She developed intermittent bleeding per vaginum which continued for another two months. Ultrasonography of pelvic region was within normal limits.

No record of beta human chorionic gonadotrophin hormone $(\beta\text{-hCG})$ estimation was present. Patient gradually developed malaise, gastritis, loss of appetite, and nausea for which she took treatment from local medical practitioner, but not benefited. On the day of admission general condition of the patient was poor, BP was not recordable, pulse rate was aprox.60 /pm., and abdomen was tender. There was no active bleeding at the time of admission. The patient had one attack of seizure during clinical examination. Paracentesis showed frank blood. Urine test for pregnancy was positive. Hemoglobin was 6.6 gm/dl. USG was not performed at that time. Other basic investigations were within normal limits.

Clinical diagnosis of ruptured ectopic pregnancy was made and emergency laprotomy was done. Intra operative findings were gross hemoperitonium, large ulceration at the posterior aspect of the uterus which was not communicating with the uterine cavity, evidence of necrosis and grape like vesicles in the pelvic cavity. Total abdominal hysterectomy with bilateral salpingectomy was done and 2 specimens were sent for HPE. Uterus, cervix, one attached fallopian tube and stump of another fallopian tube were identified in 1st specimen. On gross examination there was ulceration of 3x3x1.4 cm size present on the posterior aspect of uterus which was covered with hemorrhagic material.

There was no communication between the ulceration site and uterine cavity. Endometrium, adjacent myometrium, cervix, fallopian tube and stump of fallopian tube were grossly unremarkable. 2nd container consists of multiple grapes like structure along with some fibrous soft tissue parts. On microscopic examination the endometrium secretary, hyper secretary glands showed pseudodecidualization of stroma. Myometrium adjacent to endometrium was unremarkable. Eroded area showed infiltration of myometrium by clusters of cytotrophoblasts with prominent cytological atypia and interspersed

Figure 1: Clusters of malignant cytotrophoblasts and interspersed syncytiotrophoblasts200x

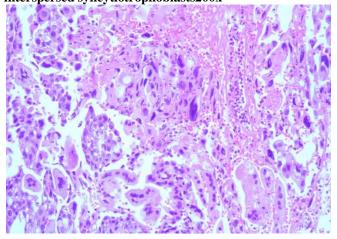
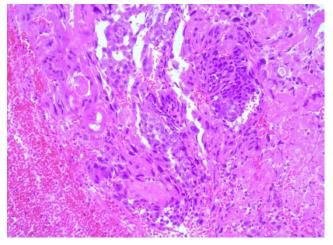


Figure 2: Infiltration of myometrium by clusters of cytotrophoblasts with prominent cytological atypia and interspersed syncytiotrophoblasts with area of necrosis and haemorrage100x



DISCUSSION

Hydatidiform mole is an abnormal conceptus, resulting due to abnormal fertilization. Based on morphological, pathological, and genetic differences it is sub-categorized into complete and partial moles .The chromosomal pattern syncytiotrophoblasts (figure1) Area of necrosis and hemorrhage were also seen (figure2) No molar villi seen in the section taken from ulceration site on uterus. Microscopic examination of second specimen showed a fallopian tube segment revealing oedema, congested blood vessel and neutrophilic infiltrate in the muscle layer (figure3).

Mucosa was unremarkable. It also showed oedematous chorionic villi with circumferential trophoblastic proliferation and cistern formation (figure 4). Surrounding tissue revealed hemorrhage and foci of necrosis. Final diagnosis was given as extra uterine (ectopic) complete mole transformed into choriocarcinoma. Post operative B-HCG was done and found to be 67400IU. CT lung showed metastatic deposits in both the lung. Follow up B-HCG showed decreasing titer.

Figure 3: Part of 2nd specimen showing fallopian tube segment revealing oedema, congested blood vessel and neutrophilic infiltrate in the muscle layer.100x

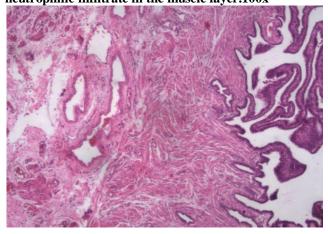
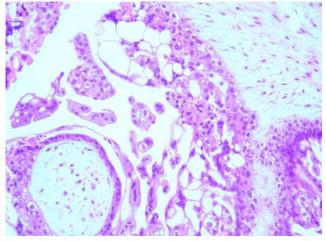


Figure 4: Part of 2nd specimen Oedematous chorionic villi with circumferential trophoblastic proliferation and cistern formation 400x.



of complete mole, is 46, XX with the genome paternal in origin[6]. The incidence of hydatidiform moles is highest in South East Asian countries. The overall incidence rate

comes to be 1 per 1,000 pregnancies [7]. Advance maternal age is the most consistent risk factor for hydatidiform mole.

Histologically, molar pregnancy is characterized by circumferential trophoblastic proliferation, hydropic changes, scalloped villi and stromal karyorrhexis [8]. The incidence of the ectopic molar gestation is very rare. Only 132 cases of Tubal ectopic hydatidiform mole and a few cases from other extra uterine site have been reported in the literature [9]. Its malignant potential is similar to that of an intrauterine molar pregnancy [10].

Choriocarcinoma is a highly malignant tumor of trophoblastic tissue. Majority of them arise due to malignant transformation of complete molar pregnancy [6] as evident in the present case. The criteria for primary extra uterine origin of Choriocarcinoma are (1) absence of disease in uterine cavity; (2) pathologic confirmation of diagnosis; (3) exclusion of molar pregnancy; and (4) exclusion of a coexistent intrauterine pregnancy [11]. This criteria was not fulfilled by our case as there was evidence of compete mole on histopathology.

The common gynecological presentation of Choriocarcinoma is abnormal vaginal bleeding and pain abdomen correlating well with present case. Feature of peritonitis due to ruptured uterus was present; Okamoto et al also reported similar case [12]. Fortunately hemoptysis, dyspnea, pleuritic chest pain, and cough were not seen in the present case despite presence of multiple patchy lesions in both lungs.

Choriocarcinoma probably one of those rare neoplasms that is completely curable by chemotherapy even in the presence of widespread metastasis. Massive life-threatening vaginal bleeding, ruptured uterus may require hysterectomy even in reproductive age women [13]. In the current case it is obvious that molar pregnancy was not suspected and no beta HCG estimation was prescribed by the medical practitioner at the time of antecedent pregnancy which resulted in the development of metastatic choriocarcinoma with ruptured uterus requiring hysterectomy. Histopathological examination remains the gold standard test for confirmation of diagnosis.

CONCLUSION

We conclude that Choriocarcinoma arising in a background of extra uterine complete mole is extremely rare condition. Ultrasonography might not be able to fully diagnose ectopic complete mole and Choriocarcinoma. There must be high degree of suspicion if abnormal bleeding PV is present in a reproductive age group female. Serum beta HCG level estimation in all suspected case and mandatory histopathological examination of the product of conception /uterine curetting in treatment unresponsive cases help to reach the early diagnosis and management of the case.

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