Recurrent Hydatidiform Mole: A Case Report of Three Consecutive Molar Pregnancies Complicated by Choriocarcinoma

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I. Introduction

Hydatidiform mole (HM) is characterized by atypical hyperplastic trophoblasts and hydropic villi. It forms a heterogeneous group of disorders, with an incidence ranging from 1 in 500 to 1 in 1 500 pregnancies in the Western World. The incidence in our region was reported to be 1 in 452 pregnancies, whereas the incidence is higher in Asia reaching 1 in 80 pregnancies[1-3]although recurrent molar pregnancies are a rare occurrence seen in 1 to 2% of cases, it is clear that women who have had a previous mole have a higher risk of recurrence than the general population[4]

We present here a case with repeated HM in three consecutive pregnancies.

II. Case Report

A 26-year-old Shabnam patient was first referred to our outpatient department (OPD) at SMS Medical College, Jaipur in January 1915 as a case of molar pregnancy for further management. She gave a history of 2 month amenorrhea, vaginal bleeding 1 day. The serum β-human chorionic gonadotropin (β-hCG) was found to be 15,444 IU and ultrasonography, multiple vesicles in endometrial cavity with bilateral adenexa normal, a chest X-ray, renal and liver function tests were normal. She underwent a uterine curettage. Histopathology reported partial hydatidiform mole.

The patient continued to have vaginal bleeding after 4 month the procedure (April 2015). The serum β-human chorionic gonadotropin (β-hCG) was found to be 9,4440 IU and ultrasonography, endometrial hyperplasis >20mm with bilateral adenexa normal ,a chest X-ray, renal and liver function tests were normal. She underwent a repeat uterine curettage in which necrotic tissue. Histopathology reported partial hydatidiform mole only.

Her subsequent clinic attendance at our hospital was irregular. The patient returned to the OPD in June, 2015 and give history to have had two more molar pregnancies treated at hospital with suction and curettage. She admitted with same complain advice ultrasonography show endometrial thickness >20mm. underwent for curettage. Histopathology report revealed avillous sheets of trophoblastic cells with hemorrhage and necrotic background. She was followed up in her base hospital in the last two pregnancies with serial quantitative β-hCG, until it became negative. By the time she came to us after 3 month of her last molar pregnancy, she complained of abdominal pain and shortness of breath. A chest X-ray showed bilateral cannon ball opacities in both lung fields which were thought to be due to metastatic choriocarcinoma; her β-hCG was 987 576.5 IU. The ultrasound of the pelvis showed an enlarged uterus (12.5 × 6.6 cm) with some cystic changes in the endometrial cavity and hypervascularity of the hypo echoic areas and intramyometrium on color Doppler. The left ovary showed three cysts, the largest was 1.7 cm. The CBC, RFT, LFT, PT, and PTT were normal.

The patient was referred to our Medical Oncology department for chemotherapy with a diagnosis of metastatic choriocarcinoma. Multiple drug regimen consisting of etoposide, methotrexate, actinomycin, cyclophosphamide, and vincristine was started. The patient required a total of 10 courses. when she was asymptomatic.

III. Discussion

The cause of molar pregnancy is unclear; however, there are several risk factors. Molar pregnancies occur at extremes of the childbearing age. For women over 40 years of age, there is a 10-fold increase, compared with only 1.3-fold increased risk in teenagers.[5]Other factors postulated to increase the risk of HM have included diet, gravidity, and contraception[6,7]

The incidence of recurrent molar pregnancy ranges from 5- to 40-fold increase in the current literature[2]In a report from the United Kingdom for women who had already had two molar pregnancies, the subsequent risk increases to 1 in 6.5 pregnancies.[4] This risk diminishes if there is a normal pregnancy following the HM.

Familial predisposition has recently been evaluated. Familial recurrent HM are considered exceedingly rare, with only 21 families reported in the medical literature. In these cases, the HM are diploid, but biparental, rather than androgenetic in origin. These patients appear to have an autosomal recessive condition, causing them to have recurrent molar pregnancies and they have very little chance of a successful pregnancy. However, this patient had no known family history of recurrent HM. Genetic studies suggest mutations in the NLRP7 gene, also known as NALP7 gene, which is located on chromosome 19q13.3–q13.4, a maternal gene, as a cause of Familial biparental HM, and possibly responsible for causing recurrent spontaneous abortions, stillbirths, and intrauterine growth retardation.[8]

Follow up of patients with HM by measuring serial β-hCG levels is very crucial to allow early detection of persistent gestational trophoblastic disease (PTD) which has high potential to malignant change. Malignant transformation may be life-threatening to the mother and needs urgent treatment. Patients with complete molar pregnancies have an increased risk of PTD, considered to be 5% compared with patients with partial molar pregnancies where it is <1%.[5]

Women who receive chemotherapy for GTD are likely to have an earlier menopause.[9] Furthermore, multiagent chemotherapy which includes etoposide increases the risk of developing secondary cancers, such as acute myeloid leukemia, colon cancer, melanoma, and breast cancer for those who survive more than 25 years.[10] These risks would necessitate long-term follow-up of these patients treated with chemotherapeutic agents

In conclusion, doctors diagnosing and managing molar pregnancies should be aware of the potential malignant transformation and the genetic predisposition for early detection and proper referral and counseling regarding the prognosis of future pregnancies.

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