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Case Report

World Journal of Medicine and Health Care

Seven Times Recurring Molar Pregnancy; Case Report and Literature Review

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Submitted: 17 September 2024 **Accepted:** 23 September 2024 **Published:** 30 September 2024

Citation: *Berhe Tesfai, Hailemichael Gebremariam, Dawit Sereke, Saron Abraham, and Okbu Frezgi (2024) Seven Times Recurring Molar Pregnancy; Case Report and Literature Review. Wor Jour of Medic and Heal Care 2(4), 01-04.*

Abstract

Recurrent molar pregnancy is not uncommon but seven times recurrences after complete cure is rare clinical entity. We report a multigravida mother managed as case of seventh episode of molar pregnancy. She had consecutive six molar pregnancies managed by suction evacuation and post molar surveillance declared cure after each pregnancy losses followed by normal pregnancy. Currently, she has seventh complete molar pregnancy, and on examination, the fundal height corresponds to 14 weeks and ultrasound revealed snowstorm appearance and urine β-hCG titer was positive at ratio of 1:4098. She was managed by suction evacuation and curettage and histopathology result revealed complete mole. Post evacuation urine β-hCG surveillance had consistently decreased and finally became negative three times in consecutive three months.

Keywords: Hydatidiform Mole, Recurrent Molar Pregnancy, β-hCG.

Introduction

Hydatidiform moles (HM) are abnormal conceptions, and small proportion of them are recurrent [1]. Recurrent molar pregnancy is repeated occurrence of molar pregnancies, which involves combinations of environmental and genetic factors [2]. Patients with a positive family history of recurrent complete moles and consanguinity are usually bi-parental and patients with a personal history of recurrent moles without family history usually has androgenetic complete hydatidiform moles [3, 4].

The risk of HM in a subsequent pregnancy increases 1–2%, 23% and nearly 100% after one, two and three consecutive molar pregnancies respectively. Of women who had a partial hydatidiform moles (PHM), 68% had another PHM and 32% complete hydatidiform moles (CHM) on another molar pregnancy [5]. The familial inheritance of the condition is also another explanation of having the same type and recurrent disease as well [6]. Ultrasound is the standard imaging modality and 'snowstorm pattern' resulting from the presence of a complex vesicular intrauterine mass containing many 'grape-like' cysts with no foetal tissue (CHM) or only partial tissue (PHM) [7].

Recurrent molar pregnancy does not always warrant chemotherapy [8]. Genetic counselling and testing, In-vitro fertilization (IVF) with intracytoplasmic sperm injection (ICSI) and preimplantation genetic diagnosis or oocyte donation may be offered to affected women Adequate follow-up of patients with HM should be done by measuring serial β-hCG levels to detect persistent gestational trophoblastic disease [9, 10]. Here, we report a patient with recurrent seven times molar pregnancy intervening with normal delivery.

Case Report

This 35-year-old, gravida 8 parity 1 mother came to Orotta National Referral Maternity Hospital with a complaint of spotting vaginal bleeding. Her last menstrual period (LMP) was on February 21st of 2023, making a gestational age of 12 weeks. She had history of consecutive six molar pregnancies at an interval of minimum a-year managed by suction evacuation and curettage. After these six recurrent molar pregnancy, she had seventh normal term delivery of the neonate with uneventful postpartum period. She had a total of eight pregnancies, with one normal term delivery and seven molar pregnancies, with five of them

were complete type. (Table: 1) The couple had no family history of similar problem, and they weren't relatives and all the pregnancies were from the same spouse. Currently, she came with complete molar pregnancy and post molar surveillance with urine β-hCG was unremarkable and cure was declared in all pregnancy losses.

Her complete blood count (CBC), chest x-ray, renal function and liver function test were unremarkable. Besides, trans-abdominal ultrasound revealed snowstorm appearance (Figure:1a) and urine β-hCG titer was positive at 1:4098. Suction evacuation and curettage was performed and the histopathology result revealed complete hydatidiform mole (Figure:1b-e). She was discharged with follow up every week through the outpatient department. On subsequent follow ups, her urine β-hCG titter was 1:128 at her first week and subsequently it became negative repeatedly for three times. Finally, she was followed monthly for three months and urine β-hCG was consistently negative and uterus was clear on ultrasound, (Figure: 1f) during which complete cure was declared.

Table 1: Timing of Pregnancies, Mode of Diagnosis and the Pregnancy Outcome

No.	Diagnosis	Pregnancy	Mode of Diagnosis	Outcome, Date
1	08/09/2011	Partial mole	Ultrasound, Histopathology, urine β-hCG	Cured, 11/06/2012
2	13/11/2013	Complete mole	Ultrasound, Histopathology, urine β-hCG	Cured, 05/ 06/2014
3	29/05/2015	Complete mole	Ultrasound, Histopathology, urine β-hCG	Cured, 24/12/2015
4	02/03/2017	Complete mole	Ultrasound, Histopathology, urine β-hCG	Cured, 17/11/2017
5	15/11/2018	Partial mole	Ultrasound, Histopathology, urine β-hCG	Cured, 18/06/2019
6	22/01/2020	Complete mole	Ultrasound, Histopathology, urine β-hCG	Cured, 07/08/2020
7	11/04/2021	Normal pregnancy	Ultrasound, Histopathology, urine β-hCG	Alive, 07/01/2022
8	21/02/2023	Complete mole	Ultrasound, Histopathology, urine β-hCG	Cured, 12/09/2023

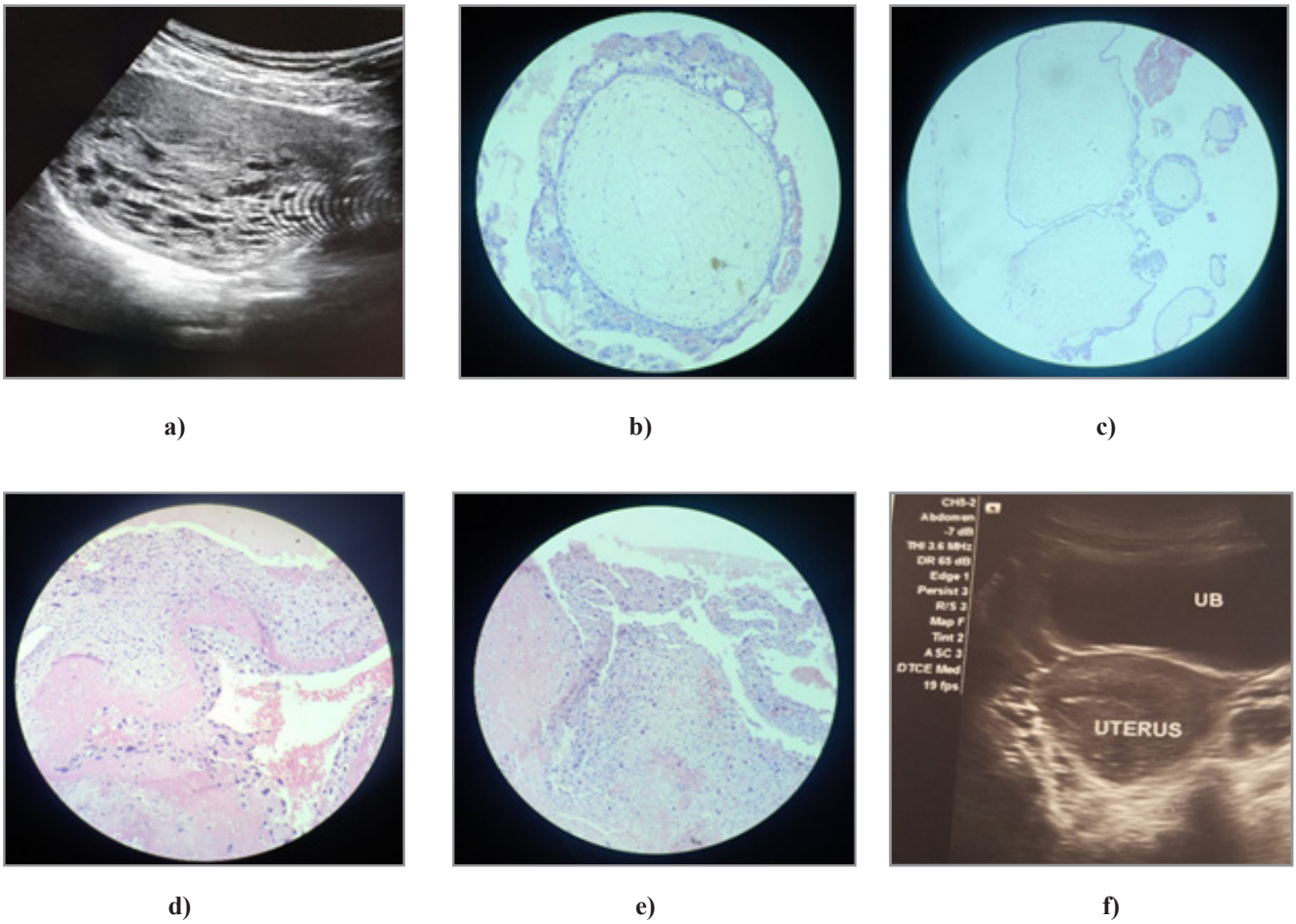

Figure 1: a) Trans-abdominal ultrasound, showed snowstorm appearance, (b-c) levels show a specimen composed and cystically dilated chorionic villi, (d-e) lined by circumscribed trophoblastic proliferation with cistern, placenta accumulation is also seen, no signs of malignancy seen on levels examined, (f) transabdominal ultrasound after post evacuation with empty uterus and clear endometrial strip.

Discussion

Recurrent molar pregnancy is associated with psychological and social challenges in the family and the incidence increases after single molar pregnancy. There are very few reports with seven consecutive molar pregnancies [11]. Regarding obstetric outcome following recurrent molar pregnancies, live birth is possible even after 4 consecutive recurrent molar pregnancies [12, 13]. Difficulty in identifying the underling etiology predisposing to recurrent mole in resource limited countries by genetic screening complicates the management of such patients.

Our case doesn't display any signs of metastasis and progression to gestational trophoblastic neoplasia during her post-molar surveillance and chemotherapy wasn't required, consistent with other studies [14]. Thus, for monitoring disease persistence, surveillance of post evacuation serial quantitative urine β-hCG levels is crucial. After these recurrent molar pregnancies, she was regularly followed for the persistence of the disease. Unfortunately, she didn't show persistence to invasive mole or gestational neoplasia in any of her molar pregnancies.

The ultrasound, histopathologic analysis and β-hCG titer results in each molar pregnancy were consistent among each other and these were consistent with other reports in literature. In patients having recurrent molar pregnancy, genetic counseling and screening are important the absence of genetic screening and karyotyping in resource limited countries is a challenge to identify underling predisposing factor. Similarly, a study reported a constrain in Sub-Sahran African countries and the evidence, which encourage genetic testing and karyotyping, as an important component of the management of affected women. Thus, incorporating of genetic testing in management of such patients could improve the outcome of this disease condition.

Based on the multiple recurrence of this molar pregnancy, early identification of the recurrence, strict management and follow-up of patients, and further investigating with genetic testing to identify the causes were among the most experience learned by clinicians in the management protocols of this disease. The case report wasn't without limitations. As genetic testing and serum β-hCG was inaccessible, this may help in the analysis and diagnosis of the case.

Conclusion

This is reported in its rarity and recurrent molar pregnancy is deeply distressing and identifying the underlying problem and counseling couples is crucial. Introduction of genetic screening and karyotyping is very important for the management of recurrent molar pregnancy in resource limited countries.

Declarations

- **Acknowledgments:** Authors acknowledges Orotta National Referral Maternity Hospital staff for management of the case.
- **Informed Consent:** Written informed consent was obtained from the patient to publish the case report for teaching purpose.
- **Funding:** This case report had no any source of funding
- **Conflict of Interest:** Authors have no any conflict of interest to disclose
- **Availability of Data and Material:** All available information is included in the manuscript.

Authors Contribution

- **Conceptualization:** Berhe Tesfai, Okbu Frezgi, Hailemichael Gebremariam
- **Writing – Original Draft:** Berhe Tesfai, Okbu Frezgi
- **Writing – Review & Editing:** Berhe Tesfai, Hailemichael Gebremariam, Okbu Frezgi, Dawit Sereke, Saron Abraham

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