Int J Cancer Manag. 2022 July; 15(7):e116981. doi: 10.5812/ijcm-116981.

Published online 2022 September 11. Case Report

A Case of Persistent Intrauterine Molar Pregnancy with Final Diagnosis of Heterotopic Molar Pregnancy: A Very Rare Entity

Zahra Shiravani [1, 2], Fateme Sadat Najib [1, 3], Mojgan Akbarzadeh-Jahromi [2] and Mojgan Hajisafari Tafti [4, *]

[1] Department of Obstetrics and Gynecology, Division of Oncology Gynecology, School of Medicine, Shiraz University of Medical Sciences, Shiraz, Iran
[2] Maternal-Fetal Research Center, School of Medicine, Shiraz University of Medical Sciences, Shiraz, Iran
[3] Infertility Research Center, School of Medicine, Shiraz University of Medical Sciences, Shiraz, Iran
[4] Department of Obstetrics and Gynecology, School of Medicine, Shahid Sadoughi University of Medical Sciences, Yazd, Iran

[*] *Corresponding author*: Fellowship of Gynecology Oncology, Department of Obstetrics and Gynecology, School of Medicine, Shahid Sadoughi University of Medical Sciences, Yazd, Iran. Email: dr.mhajisafari@gmail.com

Received 2021 July 17; **Revised** 2022 August 01; **Accepted** 2022 August 09.

Abstract

Introduction: Gestational trophoblastic disease (GTD) includes hydatiform mole, choriocarcinoma, placental site trophoblastic tumor, and epithelial trophoblastic tumor. Also, molar pregnancy can happen as an ectopic pregnancy. The coincidence of these complicated pregnancies seems to occur extremely rarely.
Case presentation: Here, we presented a 26-year-old woman, nulli gravida with the first presentation of intrauterine complete molar pregnancy; she underwent suction curettage but was prompted to Gestational Trophoblastic Neoplasm (GTN) and she received chemotherapy. During chemotherapy, she had severe abdominal pain and underwent laparotomy, and found an ectopic molar pregnancy in the fallopian tube. Salpingectomy was done and followed up with serum human chorionic gonadotropin (hCG) level and again due to improper decrease of hCG levels, she was diagnosed as a heterotopic post-molar GTN and received methotrexate (MTX) in multiple doses, but she did not respond to MTX, so we started actionomycine-D (Act-D) for her. She was cured after receiving 5 courses of Act-D and now she is on her monthly follow-up with an hCG level.
Conclusions: It is important to notice the likelihood of ectopic molar pregnancy or a heterotopic molar pregnancy in the case of managing molar pregnancy, especially when we encounter a case's poor response to medical or surgical therapy.

Keywords: Hydatidiform Mole, Ectopic, Heterotopic Pregnancy

1. Introduction

Gestational trophoblastic disease (GTD) includes hydatiform mole, choriocarcinoma, placental site trophoblastic tumor, and epithelial trophoblastic tumor (1). Also, molar pregnancy can happen as an ectopic pregnancy in the fallopian tube, cornea, and cervix. The coincidence of these complicated pregnancy cases seems to occur extremely rarely (1). To the best of our knowledge, there is only 5 heterotopic molar pregnancy (HMP) reported in the literature; moreover, pure heterotopic molar pregnancy has been reported once in the literature (2). This case report described a patient with heterotopic pregnancy with both intrauterine and ectopic fallopian tubes considered as complete molar pregnancy.

2. Case presentation

The patient was a 26-year-old nulli gravida woman who was referred to our center with the diagnosis of gestational trophoblastic neoplasia (GTN). Serum human chorionic gonadotropin (hCG) titer was 28000 mIU/mL. She underwent suction curettage in the 8th gestational week of pregnancy. Pathology examination confirmed a complete hydatiform mole. One week later, she had a second suction curettage due to an intrauterine tissue remnant of 25 mm and an inappropriate decrease of hCG to 25000 mIU/mL. However, no tissue was attained. During the second curettage follow-up, hCG titer remained plateau, so she was referred to our oncology department. According to the GTN diagnosis, we performed P/E including vaginal examination which was normal. Complete

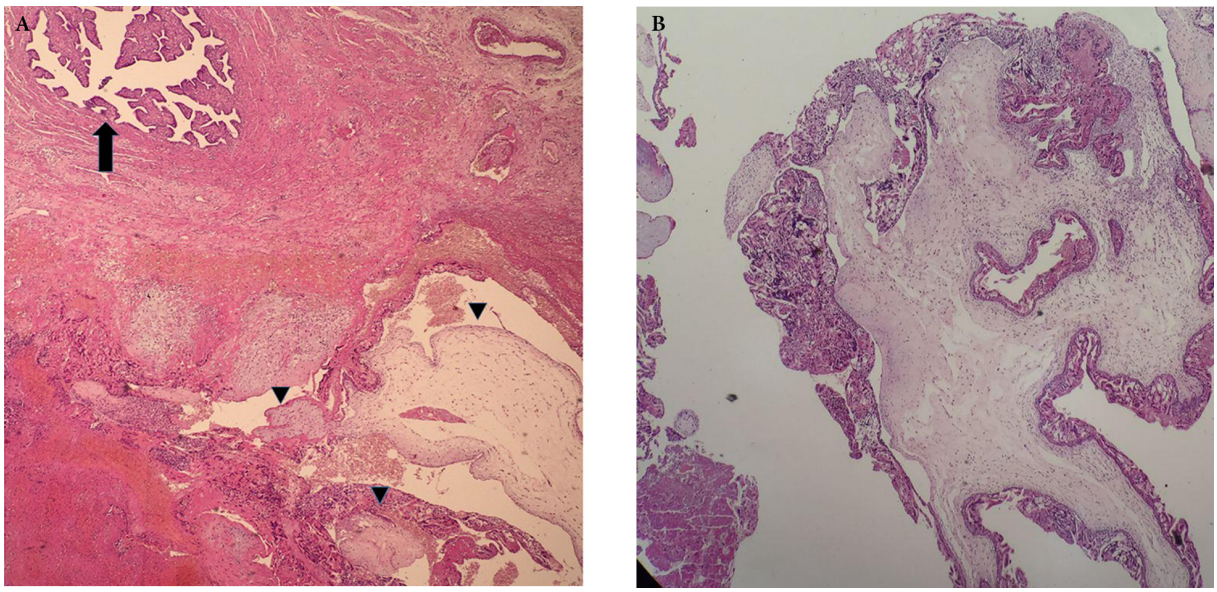

Figure 1. A, Molar tissue enlarged villi with vesicle formation and circumferential trophoblastic proliferation (H&E, ×100); B, Molar ectopic pregnancy. Fallopian tube (arrow), molar tissue (arrow head) (H&E, ×40).

diagnosed (hCG levels plateau in 4 weekly measurements over 3 weeks, rise in the hCG levels ≥ 10% in 3 measurements for 2 weeks, or abnormal hCG levels persistent over 6 months), it is necessary to repeat metastatic work-up and determine the FIGO stage and prognostic score. Metastatic work-up includes CBC differential and platelet count, liver, renal, thyroid function tests, chemistry profile, and imaging (chest, abdominal-pelvic CT scan with contrast, pelvic ultrasound or magnetic resonance imaging (MRI), and brain MRI if pulmonary metastases are present (12). Our patient was in FIGO stage I and first-line regimen was a multiday MTX regimen (1mg/kg every 2 - 3 weeks). The second line is dactinomycin in cases of initial good response to MTX which then reaches the plateau levels (12), as we prescribed for her in the second line. The limitations of our case were that it was not obvious whether GTN was due to intrauterine molar pregnancy or ectopic molar pregnancy, as both of them were complete mole. Also, Further risks of GTN in ectopic molar pregnancies were not estimated. It is so important to notice that molar pregnancy can happen as ectopic pregnancy, so if we face with poor response to medical or surgical therapy in cases of EP or even in cases of intrauterine molar pregnancy, remember it is probably an ectopic molar pregnancy or heterotopic molar pregnancy, respectively.

Acknowledgments

The authors would like to thank Shiraz University of Medical Sciences, Shiraz, Iran and also the Center for Development of Clinical Research of Nemazee Hospital and Dr. Nasrin Shokrpour for editorial assistance.

Footnotes

Authors' Contribution: Collecting data and careful fallow up: M. H., Z. Sh., F. S. N.; drafting of the manuscript: M. H.; critical revision of the manuscript for important intellectual content: Z. Sh., M. A.

Conflict of Interests: The authors declare no conflict of interests.

Data Reproducibility: No new data were created or analyzed in this study. Data sharing does not apply to this article.

Funding/Support: No grant or financial support for this study has been received by authors.

Informed Consent: The patient of this case report has been explained clearly about publishing her disease as a scientific paper without revealing her identity and she accepted. In order to publish all these data written informed consent was taken from the patient.