ISSN: 2755-984X

Journal of Tumor Science Research

SCIENTIFIC Research and Community

Case Report

Open Access

A Rare Presentation of Gestational Trophoblastic Neoplasia

Myint Myint Thinn[1*], Nwe Mar Tun[2], Kyi Thar Htun[3], Shwe Kyaw[4], Hsu Myat Mon[4], Htun Lynn Soe[4] and Thin Myat Myat Shein[4]

[1]Senior Consultant Gynaeoncologist, Central Women's Hospital, Yangon, Myanmar

[2]Professor and Head, Department of Obstetrics and Gynecology, University of Medicine (1), Yangon, Myanmar

[3]Senior Consultant Gynaeoncologist, Central Women's Hospital, Yangon, Myanmar

[4]Junior Consultant Gynaeoncologist, Central Women's Hospital, Yangon, Myanmar

***Corresponding author**
Myint Myint Thinn, Senior Consultant Gynaeoncologist, Central Women's Hospital, Yangon, Myanmar.

Received: May 08, 2023; **Accepted:** May 16, 2023; **Published:** May 24, 2023

Choriocarcinoma is an extremely malignant form of gestational trophoblastic neoplasia which usually metastasize to the lungs, liver and central nervous system. However, metastasis to the gastrointestinal tract is not a common presentation and small bowel choriocarcinoma is an extremely rare occurrence [1].

Metastatic GTN can manifest in 4% of patients after complete mole evacuation and is commonly linked to choriocarcinoma, an invasive and fast-spreading cancer type [2]. It is often difficult to determine the precise histology of choriocarcinoma. The high vascularity of trophoblastic tumors can cause hemorrhage symptoms at the site of metastasis. This, in turn, makes it difficult to obtain tissue biopsy for histology. Moreover, even though surgical intervention may help to reduce the tumour burden as well as control the massive bouts of bleeding before definitive chemotherapy which is the mainstay of treatment, it is sometimes hard for the patient to withstand the effects of double modality treatment due to the situation of the patient.

Case History

The patient was 48 years old P2+1 lady who was referred from a small township hospital for hematemesis, melena, irregular bleeding per vagina off and on with an ultrasound diagnosis of uterine sarcoma.

Her last pregnancy was ended as a molar pregnancy two years ago. Suction curettage was done and histology revealed a complete mole. She came for follow up 4 times after evacuation but never showed up again for almost two years while she was completely amenorrhoeic without any obvious gynecological problems. Two months prior to admission she started to experience irregular bleeding per vagina with mild pain and discomfort in suprapubic area. She went again to township hospital when her bleeding was profuse and became dyspneic on exertion. She was admitted for diagnostic workout there. In the meantime, she also had two massive bouts of melena stool for which she needed three packs of blood transfusion. Urine for HCG was found to be positive and sonographic examination in township hospital reported as uterine sarcoma.

She was referred to tertiary care hospital for further managements. On arrival to emergency department, she was markedly anemic with impending shock since there had been two more bouts of melena stool on the way to our hospital. On examination, there were fine crepitation and wheezing in both lung fields. Abdomen was soft and liver and spleen were not palpable. Emergency medical officer also noted that the patient was persistently coughing and brought up some blood stained sputum which was previously never complained in township hospital.

Enlarged uterus of 14 weeks' pregnancy size was also noted in abdomen. There were no vaginal metastatic nodules but bilateral cystic adnexa mases were felt along with enlarged uterus. Active bleeding through the cervix was also noted at that time. Two packs of blood were given on admission to correct anemia and revive the shock.

Serum βHCG was 144,889.00 mIU/ml on admission and blood for complete picture showed Hb of 8.5 gm/dl, WBC of 14 x 109 per litre and platelet count of 299 x 109 per litre. Other coagulation parameters were within normal limits at the time of admission. CT scan and MRI revealed choriocarcinoma of uterus with tumor size of 12x 6x 8 cm, cerebral metastasis of 1.5x 1 cm and 0.5 x 0.4 cm in right hilar region and multiple pulmonary metastasis and bowel wall thickening of 9mm with luminal narrowing with suspicious metastasis.

On second day of admission the patient got uncontrolled profuse GI bleeding and shock after massive bouts of both hematemesis and melena including fresh blood clots. She was transferred to intensive care unit. In spite of various attempts to control the GI hemorrhage, she continued to have huge bouts of bleeding

requiring massive blood transfusions with a total of 25 packs in 48 hours' period. The blood parameters deteriorated with platelet count falling to 74 x109 per litre.

Owing to a very severe Covid outbreak within our hospital, there were some delays to carry out the active interventions. The endoscopy was carried out on third day of admission and found invasive lesion in second part of duodenum with active bleeding. Endoscopist attempted to stop the bleeding but was unsuccessful. She was then given continuous infusion of Telepressin 10mg/24hr for 3 days as another option and from there, the bleeding begins to decrease. She was diagnosed as a case of choriocarcinoma stage IV with ultra-high risk score of 16.

Anemia was corrected with further blood transfusion and also with PRP and FFP. On the fifth day, the amount of bleeding was dramatically reduced and induction chemotherapy with single agent methotrexate 50mg with folinic acid rescue is started. Three days from the beginning of chemotherapy, there is no more bleeding and by the end of the week after hospitalization, she had received a total of 64 packs of blood.

The patient started to experienced weakness of the lower limbs by the end of single agent chemotherapy followed by an attack of seizure which might be attributed to brain metastasis. After finishing the induction chemotherapy and the patient was stabilized, radiotherapy of 30Gy in 10 fractions was given for the brain metastasis.

After two weeks of radiotherapy, multi-agent chemotherapy with EMACO regime was started. After two cycles of chemotherapy, the general condition of the patient is markedly improved. There were no more bleeding episodes as well as seizures. Respiratory symptoms were also improved. Serum β HCG level declined to 81,275 mIU/ml and size of the uterine tumour and pulmonary metaseries were obviously reduced.

Since the patient recovered, she requested to go home for a while before starting next cycle of chemotherapy. However, in the midst of the Covid pandemic and lockdowns along with the fact that she lived in a very remote area, she could not come back on the schedule date. And so, we could contact her back only about 3 weeks later than the scheduled date for another cycle. By then, we learned that the patient has deceased two days ago with high fever of two days without any bleeding nor seizures but with some cough and respiratory symptoms. Unfortunately, we lost the patient and it was never known if the patient has died due to Covid or due to chemotherapy related complications with septicemia.

Discussion

Choriocarcinoma is a highly malignant epithelial tumor which can be associated with any type of gestational event, mainly with complete hydatidiform mole. About 2-3% of complete moles can progress to choriocarcinoma while 50% of choriocarcinoma cases are diagnosed after a molar pregnancy [2,3].

Although distant metastasis is a not very common consequence of complete molar pregnancies after complete evacuation but if it occurred, usually associated with choriocarcinoma while the precise histology is often not determined.

Commonest site of metastasis is lungs followed by vagina, pelvis, brain and liver. Metastasis to the gastrointestinal tract is quite rare along with spleen and kidney which accounts for <5% of all distant metastasis [3].

According to the case series and literature review by Wang et al, some authors stated that patients with intestinal metastases from gestational choriocarcinoma inevitably have a combination of pulmonary and hepatic metastases but in our case there were brain and pulmonary metastasis altogether with GI lesion making the treatment much more difficult [4,5].

Up to now there is no consensus on treatment of gastrointestinal metastasis of choriocarcinoma because of its rare occurrence and limited knowledge. In review of the case reports, surgical resection of the effected coils of intestine followed by multi agent chemotherapy gave some promising results [6,7]. However, in this presenting case, general condition of the patient was critically ill with brain metastasis and respiratory symptoms while having repeated bouts of GI bleeding making neither surgery nor multiagent chemotherapy in applicable.

Main reason of this case report is to highlight the treatment of a critically ill and hemodynamically unstable patient. Due to this, we could not operate on her and we also cannot start multiagent chemotherapy in the very beginning. Moreover, even though active lesion was found in second part of duodenum, the lesion from the large gut could not be identified properly owing to massive amount of blood along with melena stool during colonoscopy. Hence, as an alternative treatment, we tried continuous intravenous infusion of Novapressin (Terlipressin) and the bleeding was obviously reduced about 7-8 hours after starting the drug.

During this period, we considered that she could not bear the toxicity of the multidrug regime and so, single agent methotrexate 50mg alternate day with folinic acid rescue is given as an induction chemotherapy. The aim is to shrink the active bleeding lesion. Bleeding was satisfactorily controlled after single agent chemotherapy.

In the case report by Ramessur et al, the patients with intestinal metastases from choriocarcinoma, causing intussusception and bleeding from intestinal lesions was treated with low-dose EP chemotherapy regimen (etoposide 100 mg/m2 and cisplatin 20 mg/m2). After 6 days of the initiation of chemotherapy, recheck CT showed complete resolution of intussusception and bleeding was also reduced [8]. These findings may support the role of single agent or low dose chemotherapy for the patients who are not fit for surgery before starting definitive treatment.

After completion of radiation for brain metastasis and two cycles of EMACO, the patient showed a dramatic response with marked decrease in the lesion size of both lung and brain. Unfortunately, due to various reasons, she could not come and receive chemotherapy after 2 cycles. In the end, we found out that she passed away without knowing the exact cause of death. Though most of the case reports recommend surgery such as resection of the diseased intestine and anastomosis afterwards followed by chemotherapy, in this case, the patient was very ill and could not withstand surgery but also multiagent chemotherapy. Therefore, we had to use vasopressin analog followed by single agent chemotherapy. Only after that, we could begin the definitive managements.

Overall, even though the patient has deceased, we have chosen this case as an exemplary treatment of a difficult case with multiple complications as well as to high light the role of vasopressin analogue and induction chemotherapy for the management of major bleeding from intestinal metastatic choriocarcinoma.

References

1. Cho EB, Byun JM, Jeong DH, Yoon HK, Kim YN, et al. (2016) Metastatic choriocarcinoma as initial presentation of small bowel perforation in absence of primary uterine lesion: a case report. Tumori 102.
2. Rushdan M Noor, Tay Eng Hseon, Jeffrey Low Jen Hui (2011) Gestational trophoblastic diseases. In: Handbook of gynaecology oncology for specialists and trainees.Second Edition. PerpustakaanNegara Malaysia https://koha1.imu.edu.my/cgi-bin/koha/opac-detail.pl?biblionumber=39130.
3. Berkowitz RS, Goldstein DP, JS Berek, NF Hacker (2015) Gynecologic Oncology. 6th edn. USA, Wolters Kluwer file:///C:/Users/User/Desktop/Jonathan%20S.%20%20Berek.pdf.
4. Wang Y, Wang Z, Zhu X, Wan Q, Han P, et al. (2022) Intestinal metastasis from choriocarcinoma: a case series and literature review. World J Surg Oncol 20: 173.
5. Ravi B, Dalal AK, Sharma U, Dhall JC (1997) Choriocarcinoma of the small gut. Acta Obstet Gynecol Scand 76: 712-713.
6. Balagopal PG, Pandey M, Chandramohan K, Somanathan T, Kumar A (2003) Unusual presentation of choriocarcinoma. World J Surg Oncol 1: 4.
7. Fatemi SR, Hatami B, Ghobakhlou M, Mohsenifar Z (2017) Severe lower gastrointestinal bleeding due to metastatic choriocarcinoma to the jejunum: a rare presentation. J Gastroenterol Dig Dis 2: 22-24.
8. Ramessur A, Openshaw M, Sarwar N (2015) Intussusception in gestational choriocarcinoma (not histologically proven), resolving spontaneously with chemotherapy. BMJ Case Rep 2015: 23-24.