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Case Report

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Fertility Preservation Approach in a Case of Perforating Gestational Trophoblastic Neoplasia

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ABSTRACT

Background: Gestational trophoblastic neoplasia following spontaneous abortion is rare and in majority cases the approach is hysterectomy followed by chemotherapy.

Case: We present case of a 22-year-old woman who presented with complaints of vaginal bleeding for 2 months; pain abdomen for 3-4 days. On reviewing history, patient was one and a half months pregnant and had spontaneous abortion for which she underwent twice dilatation and curettage and once laparotomy in outside hospital. Patient continued to bleed & MRI pelvis was done which revealed arteriovenous malformation and was thereafter referred to our hospital. On presentation her vitals were stable with pallor positive. On per-abdominal examination there was generalized tenderness and mild bleeding on per speculum examination. Her serum beta hCG at the time of admission was 98,156IU/L. Ultrasound pelvis revealed a highly vascular heterogenous hypoechoic mass showing significant internal and peripheral vascularity. We decided for uterine artery embolization owing to highly vascular GTN, young age and no live issues with the aim of uterine preservation. This was followed by chemotherapy with serial beta hcg monitoring. She is in her 14th month of follow up with resolution of her complaints.

Conclusion: The aim behind presenting this case is its rarity of presentation as well as management to increase awareness among treating gynecologists. Importance of multi-disciplinary approach should not be undermined in challenging cases.

Keywords: Gestational Trophoblastic Neoplasia, Uterine Artery Embolization, Uterine Preservation, Spontaneous Abortion

Introduction

Gestational trophoblastic neoplasias are characterized by aggressive invasion of the endometrium and myometrium of the uterus by trophoblastic cells [1]. It includes a spectrum of disease from benign hydatidiform mole to malignant gestational trophoblastic tumor (invasive mole, choriocarcinoma, and placental site trophoblastic tumor. Persistent gestational trophoblastic neoplasia is evidenced by the persistence of trophoblastic activity following evacuation of molar pregnancy or following previous abortion or ectopic pregnancy or even normal pregnancy. Gestational trophoblastic neoplasia follows hydatidiform mole in 60% cases, previous spontaneous abortion in 30% and normal pregnancy or ectopic gestation in 10% cases [2,3].

GTN usually presents with vaginal bleeding, soft and enlarged uterus and persistently raised beta hCG levels [1].

Case Report

A 22-year-old woman came to us referred from other hospital in view of severe anemia and possibility of arterio-venous malformation (AVM). On enquiring from the patient, she told she had history of amenorrhea for one and a half months, three-months back. She got her urine pregnancy test done, which was positive and did not get any ultrasound done. She started having spontaneous bleeding per vaginum and went to a private clinic nearby where she underwent surgical evacuation and was discharged in a stable condition. Nearly, 10 days following evacuation, she started having pain abdomen and bleeding per vaginum for which she went to another hospital where an ultrasound was done and findings were suggestive of ruptured ectopic. She was then subjected to laparotomy which as per patient's husband was negative laparotomy with no adnexal mass. On post-operative day 6, patient again started bleeding per vaginum and ultrasound revealed vascular retained products of conception. She was again subjected to curettage. All throughout her hospital stay, she received 3 units of packed cell volume and 4 units of fresh frozen plasma. She was discharged following curretage. Following discharge, patient started having abdominal

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distension with inability to pass motions, fever and bleeding per vaginum continued. She went back to the treating hospital where she underwent MRI-pelvis following stabilization and thereafter referred to our hospital owing to the possibility of AVM and sick condition of the patient. This was her first conception with no significant past or family history.

On examination, she was febrile, pale looking, and dehydrated. RT (Ryle's tube) in situ had 500 cc of bilious contents. Her pulse rate was 92 beats per minute, blood pressure was 100/60 mmHg, respiratory rate was 26 per minute, saturation was 98% on room air and temperature was 990F. Her chest examination revealed bilateral infrascapular crepts. Her cardiovascular examination was normal. On per-abdominal examination, a transverse scar was seen two-finger breadth above symphysis pubis. There was generalized tenderness, guarding and rigidity. Bowel sounds were present. On per-speculum examination, cervix and vagina were normal looking with bleeding per vaginum. On per-vaginal examination, uterus was bulky with bilateral fornices free. On per-rectal examination, same findings were confirmed.

Her baseline investigations were hemoglobin 6.9 gm%, platelet count 84000/mm3, INR 2.31. Her liver and kidney function tests were normal. Chest X-ray revealed patchy areas of haziness in bilateral perihilar region with prominent right hilar shadow, bilateral CP angles clear. Patient had MRI-Pelvis done prior to presenting to us. The findings were of a large ill-defined irregular signal intensity mass of size 5x6x5.4cm in anterior wall of fundus and body of uterus with tortuous serpiginous irregular vascular channel characterized by flow void and vascular channel involving posterior wall of uterine body and fundus with focal contour bulge anteriorly. Anteriorly lesion reached upto outer surface of myometrium with focal irregularity of anterior myometrium with multiple tortuous flow voids in bilateral parametrium.

Ultrasound pelvis done in our hospital revealed a highly vascular heterogeneous hypo echoic mass of 5.8cmX5.5cmX6.5cm in the region of fundus and anterior myometrium showing significant internal and peripheral vascularity (figure 1) and displacing endometrium posteriorly. Moderate free fluid was found without breach of uterus. Bilateral adnexa were normal looking. Ultrasound guided tap was done and revealed hemoperitoneum. Her baseline beta hCG at the time of presentation was 98,156mIU/L. Owing to high beta hCG values and radiological images suggestive of vascular invading mass, diagnosis of GTN was made.

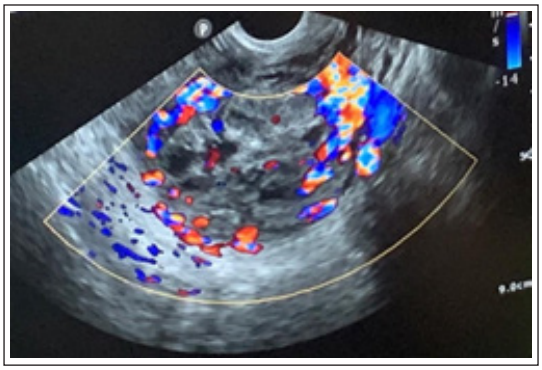

Figure 1: Pelvic USG - Doppler

Owing to the poor general condition of the patient; nulligravida; young age and high possibility of hysterectomy if evacuation is done for histopathological diagnosis, we decided for selective uterine artery embolization after optimization in consultation with intervention radiologist and medical oncologist. She was given 1 unit PCV and 4 units of FFP's prior to uterine artery embolization. CT angiography done at the time of embolization revealed bulky uterus with multiple tortuous dilated arterial and venous channels in anterior myometrium and bilateral parametrium as seen in figure 2.

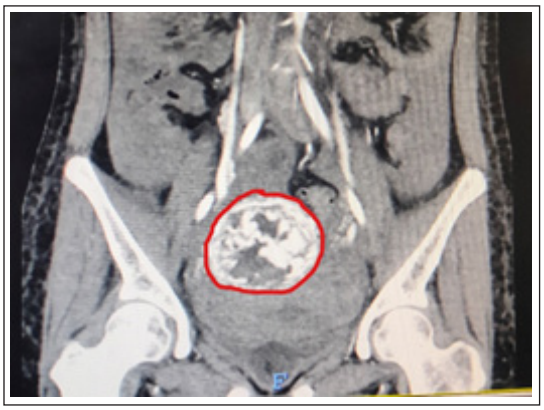

Figure 2: CT Angiography showing bulky uterus with multiple dilated tortuous channels

In consultation with oncologists, she was started on single dose methotrexate. Ultrasound pelvis at the time of discharge revealed no free fluid following UAE and first cycle of chemotherapy. She received total 5 cycles of single dose methotrexate and thereafter four cycles of EMACO regimen due to rise in beta hCG levels as can be appreciated in line diagram (figure 3). She was followed up with beta hcg values weekly till normal. She resumed with her normal menstruation.

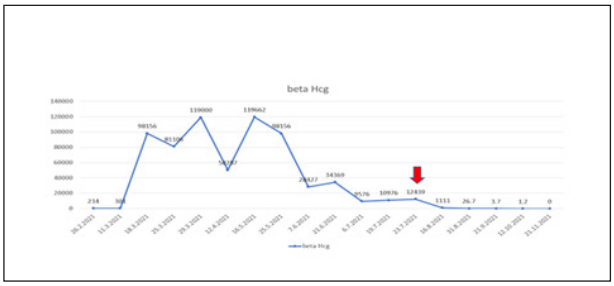

Figure 3: Line diagram showing beta hCG trend. Arrow head shows beta hCG value at which EMACO was started.

Patient has been in follow up with us and is doing fine.

Discussion

Gestational trophoblastic neoplasia arises when normal regulatory mechanism controlling the proliferation and invasion of trophoblastic tissue is lost. GTN are rare tumors that constitute less than 1 % of all gynecological malignancies [4]. The diagnosis of GTN is made on the basis of elevated beta hCG levels supported, if possible, by histopathological diagnosis of choriocarcinoma or radiological evidence or presence of metastasis [5,6]. In our case, it was high levels of beta hCG along with radiological evidence that helped us in making the diagnosis. In our case there had been increase in beta hCG of 325% over period of two week and serum hCG had increased more than 20,000 after four weeks of evacuation.

On reviewing the literature, it was found that hysterectomy was main stay of treatment in majority of such cases.

Budiana et al reported an atypical presentation of invasive mole. A 40-years-old P_1L_0 woman presented with 9 months amenorrhea with negative urine pregnancy test. Ultrasound pelvis was suggestive of neoplasia with beta hCG of 55.8mIU/ml. This patient was subjected to curettage after two cycles of methotrexate and it led to profuse bleeding. She was stabilized and further cycles completed. But on follow up, due to enlarging uterine size despite normal beta hCG she was subjected to hysterectomy [7]. The authors concluded that invasive mole can be treated in presence of clinical and radiological diagnosis in absence of pathological examination. Similarly, we also made a clinical and radiological diagnosis and used uterine artery embolization to cater to active oozing that was going on due to vascular mass that led to hemoperitoneum followed by chemotherapy [7].

In a case by S. Ahmed et al, a 25-years-old, $P_2L_2M_1$ woman presented with complaints of severe lower abdominal pain, vomiting with history of fainting attack and bleeding per vaginum. She had a history of suction evacuation and curettage for molar pregnancy 4 months back which was confirmed as hydatiform mole with no malignancy. Ultrasound abdomen revealed massive hem peritoneum and she was taken for laparotomy. Intraoperative findings had multiple perforations in uterus and bleeding was present from all perforated sites and patient underwent hysterectomy [8]. Our case was also a case of perforating invasive mole which was the reason for hemoperitoneum at presentation. But, uterine artery embolization was a boon to prevent hysterectomy in our case.

Conclusion

Invasive mole is the most common form of gestational trophoblastic neoplasia. It can be diagnosed clinically, and treatment plan consists of chemotherapy and / or surgery which can be initiated in the absence of pathological examination. Importance of involving senior gynecologist, intervention radiologists and medical oncologists to optimize the outcome in such cases are of utmost importance. Such panoramic approach helps in decreasing morbidity of patients. Also, it is important to consider either getting a scan in case of spontaneous abortions prior to proceeding for evacuation or send the tissue retrieved for histopathology. This simple step can prevent patients from multiple interventions and timely management.

Conflict of Interest: None

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