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Case Report

Role of surgical management in invasive mole: a report of 2 cases and review of literature

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ABSTRACT

Invasive mole is a rare gestational trophoblastic neoplasia with proliferative trophoblast invading into myometrium or uterine vasculature. Primary management of invasive mole is chemotherapy, but hysterectomy can be performed in selective cases. In this report, we discuss two cases of invasive mole, which required surgical intervention in the form of a hysterectomy. Both patients had a favorable outcome and are in remission.

Keywords: Surgical management, Beta hCG, Hysterectomy, Chemotherapy, Invasive mole

INTRODUCTION

Gestational trophoblastic disease (GTD), a group of pregnancy-related tumors, is characterized by abnormal proliferation of the trophoblast layer of the placenta. It can be divided into four clinicopathologic forms: hydatidiform mole (complete and partial), invasive mole, choriocarcinoma, and placental site trophoblastic tumor (PSTT)/epitheloid trophoblastic tumor (ETT).[1,2] Hydatidiform mole is benign, and the rest are malignant. Chemotherapy is the primary mode of treatment in choriocarcinoma and invasive mole, while surgery is the mainstay of treatment in cases of PSTT.

Invasive mole is a malignant trophoblastic disease with proliferative trophoblast invading into the myometrium or uterine vasculature. It usually presents in reproductive-age women.[3,4] The incidence of an invasive mole is 1 in 15000 pregnancies. The diagnosis of an invasive mole is usually made on histopathology (HPE), but with the advent of imaging modalities and beta hCG values, it can be diagnosed beforehand. Chemotherapy is the first line of management. The role of hysterectomy is limited in such chemosensitive tumors. However, it must be performed in a few patients with acute abdomen, heavy vaginal bleeding, or sepsis.

We present two such cases where we had to perform a hysterectomy in patients with suspected invasive mole, and the outcome was favorable in both cases.

CASE REPORT

Case 1

A 38-year-old female, para 3, live 3, last childbirth seven years back, presented with a complaint of amenorrhoea for three months, gradually increasing mass per abdomen, and vaginal bleeding for 15 days. Her general physical examination was within normal limits except for moderate pallor. The uterus was 20 weeks in size on abdominal examination, firm in consistency, smooth surface, side-to-side mobility present, and non-tender. Cervix appeared bulky with prominent vessels and brownish dirty discharge seen coming through the cervical os; ~20 weeks uterine mass was felt, bilateral fornices were free and non-tender, and no nodularity was felt in the pouch of Douglas on vaginal examination.

Her urine pregnancy test (UPT) was positive. Transvaginal sonography (TVS) was suggestive of complete molar pregnancy with a hyperechoic area (11×9.7 cm) in the endometrial cavity. Hemoglobin value was 7.8 g/dl, thyroid stimulating hormone (TSH) was 0.006 micro-IU/ml, and beta hCG report was >2 lakhs. A metastatic workup was performed in view of the suspicion of a high-risk mole. Chest X-ray was normal. Magnetic resonance imaging (MRI) pelvis showed an endometrial cavity lesion measuring 9.3×9×12 cm with multiple cystic spaces and diffuse thinning of the myometrial wall, suggestive of an invasive mole (Figure 1).

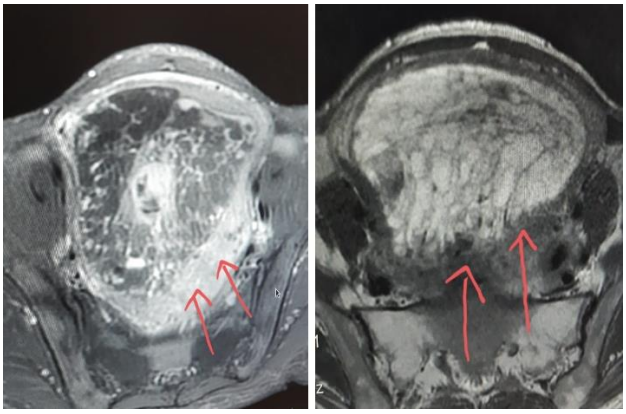

Figure 1: T1 and T2 weighted MRI images showing myometrial invasion (axial view).

During her hospital stay, 2 PRBC units were transfused, and sought an endocrine opinion for hyperthyroidism. WHO prognostication risk assessment score was 12, suggestive of high-risk GTN. The case was discussed in the institutional tumor board and planned for multi-agent chemotherapy (EMA-CO). However, an emergency hysterectomy was performed because of persistent vaginal bleeding and the patient's unwillingness for primary chemotherapy. Figure 2 shows the gross uterine specimen and cavity filled with grapes-like vesicles and myometrial thinning. Histopathology confirmed the diagnosis of an invasive mole (Figure 3).

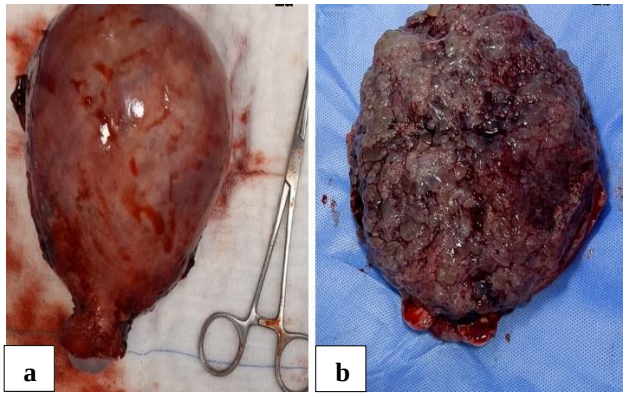

Figure 2: (a) Gross uterine specimen; (b) cut section shows uterine cavity filled with grape-like vesicles and myometrial thinning.

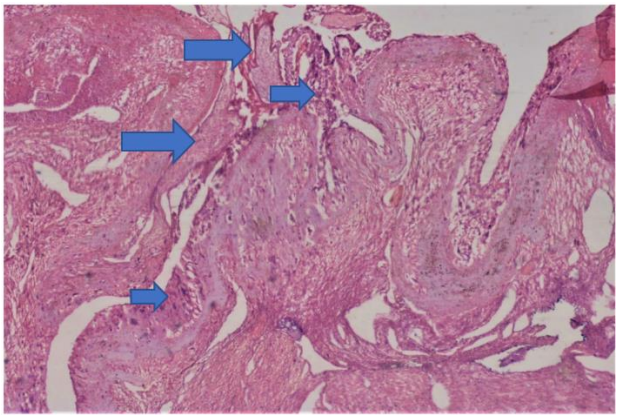

Figure 3: H&E stained (200x) image shows invading villi in the myometrium and trophoblast proliferation.

Beta hCG after 48 hours was 3500 IU/l, followed by weekly beta hCG monitoring was done till three negative values. Figure 4 shows a weekly trend of beta hCG. The patient was compliant yet refused adjuvant chemotherapy. Currently, she is under strict vigilance and monthly beta hCG monitoring for 5 years.

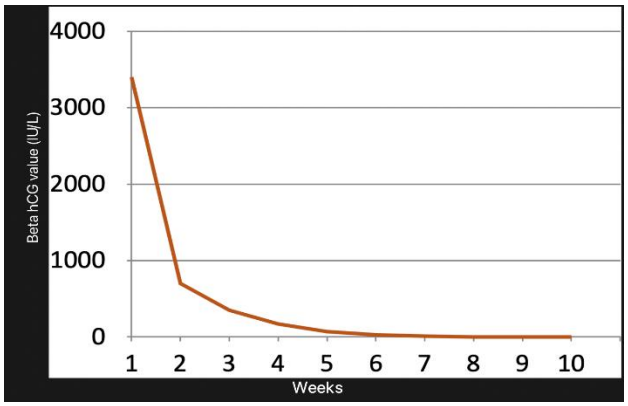

Figure 4: Weekly beta hCG follow-up.

Case 2

A 51-year-old perimenopausal female para 3, live 3, abortion 1, last conception was in the form of spontaneous abortion 16 years ago, presented with continuous vaginal bleeding for three months and abdominal pain. Her UPT was positive, beta hCG was >2 lakhs IU/l, and TVS showed a bulky uterus with a large heteroechoic lesion (7.2×9.4×11.7 cm) with variably sized cysts within filling the endometrial cavity with indistinct fat planes with the myometrium, likely GTD. MRI showed a heterogenous endometrial lesion measuring 11×10×13 cm, causing myometrium thinning suggestive of an invasive mole. Similar to the first case, tumor board discussion was done in view of high-risk GTN (WHO score-12) and planned for multi-agent therapy (EMA-CO). During the first cycle of chemotherapy, EMA was given on day 1 and 2. On day 6, the patient developed fever and neutropenia (total leucocyte count-1000/microliters), which was managed

conservatively, and the CO regimen on day eight was withheld.

The patient developed heavy vaginal bleeding, the passage of grape-like vesicles, and severe abdominal pain on day nine and was taken up for emergency total abdominal hysterectomy with B/L salpingo-oopherectomy. Figure 5 shows the intraoperative uterine specimen and around 500 gm grape-like vesicle mass removed through the vagina. Two PRBCs were transfused intraoperatively. Her postoperative course was uneventful. HPE reported an invasive mole, and the patient was planned for three cycles of EMA-CO regimen, followed by beta HCG monitoring weekly till three negative values and monthly for five years. After two months, her beta hCG value was negative, and the ultrasound abdomen and pelvis were normal.

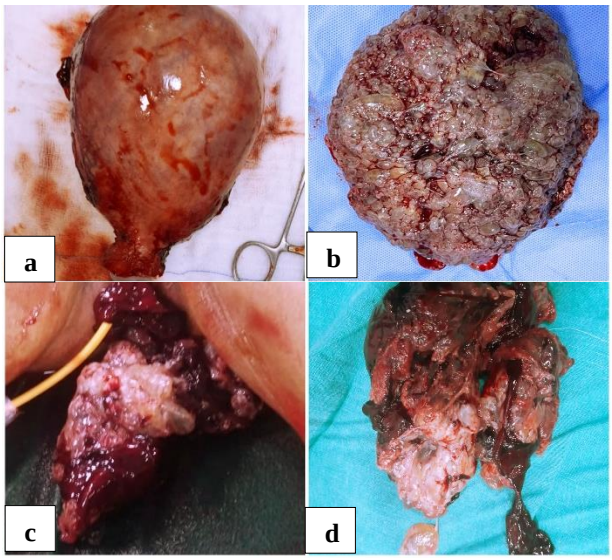

Figure 5: (a) Gross uterine specimen; (b) cut section shows uterine cavity filled with grape-like vesicles and myometrial thinning; (c) and (d) protrusion of vesicular mass through vaginal introitus.

DISCUSSION

The first line of management of invasive mole is chemotherapy, according to the WHO prognostication score.[5] The role of surgical management is limited in women with non-metastatic GTN, older patients with completed family, non-compliant patients, unable to follow up and not willing to chemotherapy, and in emergency situations e.g. acute abdomen, major hemorrhage, or sepsis. Hysterectomy decreases the tumor load and need for multiple courses of chemotherapy. Additionally, it can be considered as an adjuvant treatment in patients with chemoresistance and increased risk of recurrent GTD.[6-9]

In a retrospective data analysis from 1996-2006, Cagayan et al advocated that early surgical intervention along with chemotherapy can reduce the number of cycles needed to attain remission and decrease the chemotherapy-induced toxicity.[10] Post-chemotherapy, the incidence of adverse drug reactions is 21.5%, and the probability of chemoresistance is 20-40%.[6] Likewise, Clark et al in their retrospective study (1959-2009) reported a remission rate of 84.7% in patients with hysterectomy for GTN. Moreover, patients who underwent hysterectomy for chemotherapy-resistant disease, 75.8% subsequently achieved complete remission.[11] Therefore, hysterectomy plays a vital role in the management of selected patients with GTN.

A strict follow-up with weekly beta hCG till 3 negative values, then monthly for five years to be done even after a hysterectomy. In case of rising or persisting beta hCG, chemotherapy should be given.

CONCLUSION

Invasive mole is a rare variety of gestational trophoblastic disease. The first line of management is chemotherapy. The role of hysterectomy is limited in cases of chemoresistance, major hemorrhage, sepsis, and patient's unwillingness. The advantages of hysterectomy are avoidance of chemotherapy-induced toxicity and reduced risk of recurrence and morbidities. Hysterectomy plays a vital role in the management of invasive mole as either a primary/adjuvant or emergency procedure.

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