Surgical Approach for Uterine Perforation due to Gestational Trophoblastic Neoplasia: A Case Report

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Abstract

Introduction: The most common life-threatening complication of gestational trophoblastic neoplasia (GTN) is uterine perforation. In several cases, a surgical approach becomes inevitable. However, there are numerous considerations regarding the technique of hysterectomy.

Case Presentation: A 37-year-old (P1A2) woman came to the ER with severe abdominal pain 12 h before admission. The patient had been previously diagnosed with GTN and was scheduled to undergo methotrexate chemotherapy. However, due to the development of an acute abdomen, accompanied by low hemoglobin levels and free fluid showing signs of uterine perforation, an emergency total abdominal hysterectomy was performed. The intra-operative findings revealed intra-abdominal bleeding and uterine perforation at the right fundal side of the uterus. The patient underwent postoperative chemotherapy and showed significant improvement.

Conclusion: Regardless of the efficacy of chemotherapy in GTN, abdominal hysterectomy proved to be beneficial for treating uterine perforation in an emergency setting.

Keywords: Uterine perforation, Trophoblastic neoplasia, and Hysterectomy

Pendekatan Pembedahan pada Perforasi Uterus Akibat Tumor Trofoblastik Gestasional : Sebuah Laporan Kasus

Abstrak

Pendahuluan: Komplikasi paling umum yang mengancam jiwa dari Tumor Trofoblastik Gestasional (TTG) adalah perforasi uterus. Pendekatan pembedahan tidak dapat dihindari dalam beberapa kasus. Namun, ada banyak pertimbangan mengenai teknik pembedahan dalam setiap kasus.

Laporan kasus: Seorang wanita berusia 37 tahun (P1A2) datang ke UGD dengan nyeri perut hebat 12 jam sebelum masuk rumah sakit. Pasien didiagnosis dengan neoplasia trofoblastik gestasional dan dijadwalkan untuk kemoterapi metotreksat. Abdomen akut, kadar hemoglobin rendah, cairan bebas menunjukkan tanda perforasi uterus. Temuan intraoperatif adalah perdarahan intra-abdomen dan perforasi uterus di sisi fundus kanan uterus. Histerektomi abdomen total darurat dilakukan. Pasien menjalani kemoterapi pasca-operasi dengan kondisi yang membaik.

Kesimpulan:Terlepas dari efektivitas kemoterapi pada TTG, histerektomi abdominal bermanfaat untuk mengobati perforasi uterus dalam keadaan darurat.

Kata kunci: Histerektomi, Perforasi uterus, Trofoblastik neoplasia

Indonesian Journal of Obstetrics & Gynecology Science is licensed under a Creative Commons Attribution-ShareAlike 4.0 International License (https://creativecommons.org/licenses/by-sa/4.0/) **DOI:** 10.24198/obgynia.v8i1.776

Introduction

The term "gestational trophoblastic disease" (GTD) refers to a group of pregnancy-related illness caused by aberrant trophoblastic tissue proliferation. According to the literature, the incidence of GTD ranges from 1:500 to 1:1000 pregnancies. The disease spectrum includes benign disorders, including partial and complete molar pregnancies, as well as the category of malignancies is gestational trophoblastic neoplasms (GTN), consisting of invasive molars, choriocarcinoma, placental site trophoblastic tumor, and epithelioid trophoblastic tumor.[1,2]

In 75%–80% of cases, haploid (23 X) sperm fertilize the ovum, which then copies its own chromosomes to generate complete hydatidiform moles. The characteristics of this group include lack of embryonic development, a 46 XX karyotype (less frequently 46 XY), diffuse swelling of the villi, and diffuse trophoblastic hyperplasia. They are often linked with an increased risk of both local and metastatic invasion and a beta-hCG level of >100,000 IU/L. The characteristic of invasive moles is myometrial invasion that may result in uterine perforation. There were risks of uterine perforation in GTN, such as tumor size and location (larger tumors or tumors located near the serosal surface of the uterus increase the risk) and delayed treatment. In 15% of the cases, metastases were typically seen in the lungs and less frequently in the vagina.[3,4]

Until, 1956, malignant GTN was considered an incurable cancer However, Li et al. reported the first-ever full remission of GTN in a patient with metastatic choriocarcinoma. GTN is one of the most treatable gynecologic cancers because it is chemosensitive. With the current treatment course, the cure rate for GTN is 80% for metastatic disease and nearly 100% for nonmetastatic disease.

In some cases, surgical management, such as hysterectomy, is required. However, primary treatment consists of chemotherapy with single-agent (methotrexate or actinomycin) or multi-agent (etoposide, methotrexate, actinomycin, cyclophosphamide, and vincristine (EMACO)).[5,6] This report presents an emergency case of uterine rupture in a patient with GTN who had not received chemotherapy before.

Case

A 37-year-old (P1A2) woman came to the ER with severe abdominal pain 12 h before admission. The pain increased gradually within hours. There was no complaint of vaginal bleeding. The patient underwent curettage and was diagnosed with hydatidiform mole in February 2024. She was diagnosed with GTN (FIGO score 6) due to an increase of β-hCG level (151.453 mIU/mL to 204.778 mIU/mL) within 3 months post molar curettage (no histopathology result) and was scheduled for methotrexate chemotherapy.

On physical examination, tachycardia (110 beat/minute) was noted. An acute abdomen was noted based on abdominal muscular defense, pain, and shifting dullness during physical examination. The uterus was enlarged, corresponding to 12–14 weeks of gestational age. Gynecological examination revealed bulging of the douglas pouch during vaginal touching. Ultrasonography showed an anteflexed uterus measuring 8.23 × 4.75 cm, with dehiscence within the myometrial line from the uterus. An intracavitary mass with hydrophilic villi was observed, and the color score was +4. The bilateral adnexa were within normal limits. Fluid was collected during the examination (**Figure 1**).

Laboratory results showed an hb level of 8.6 mg/dl, and BhCG from time to time post molar evacuation was 151.453 mIU/mL on March 7th 2024, 64202 mIU/mL on April 5th 2024, 163.112 mIU/mL on April 24th 2024,

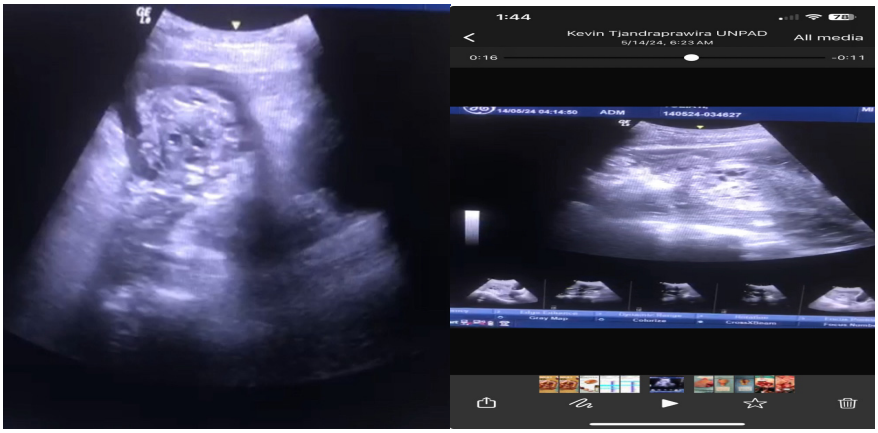


Figure 1 Myometrial dehiscence and free fluid collection

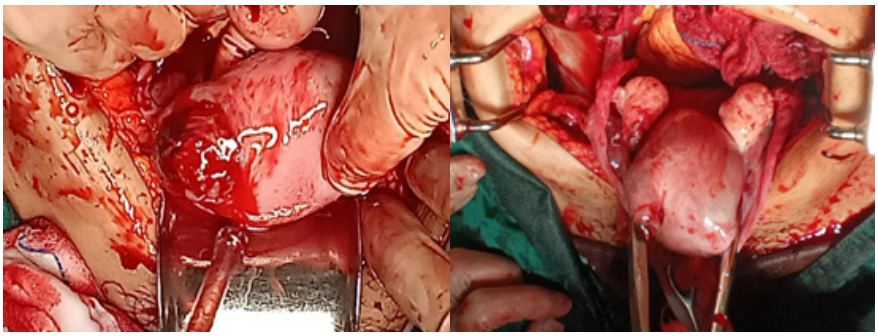


Figure 2 Intra-operative findings

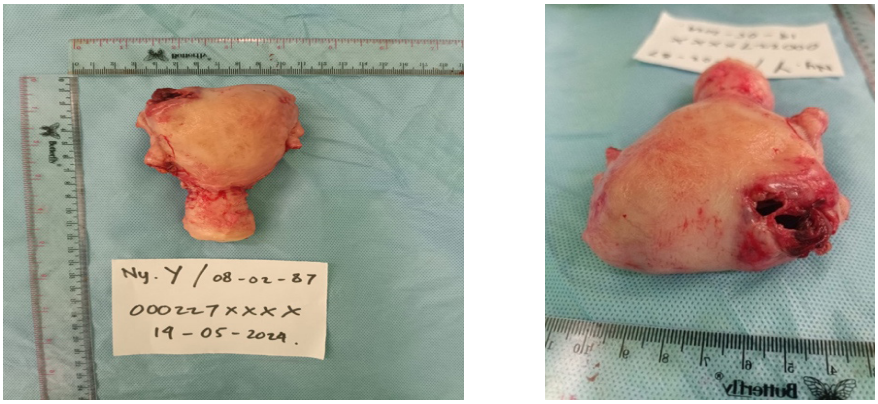


Figure 3 Histopathology specimen

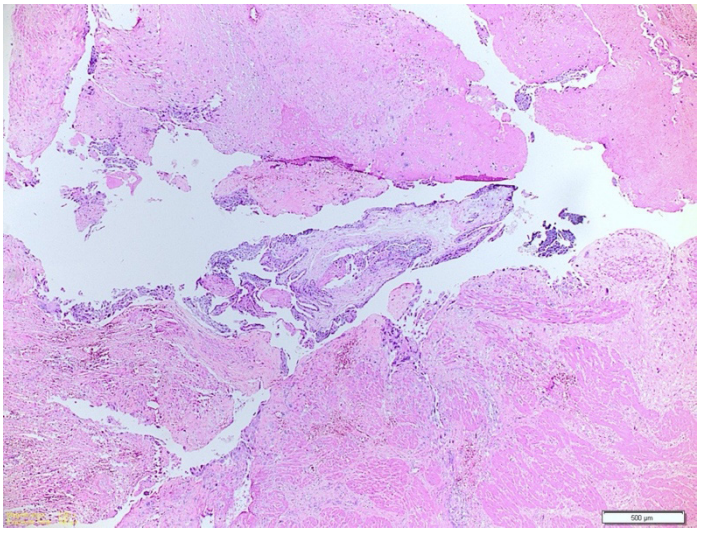


Figure 4 Villi with abnormal myometrial trophoblastic proliferation

and 204.778 mIU/mL on May 2nd 2024. During an emergency laparotomy, intra-operative findings showed blood and blood clot ±1500 cc, and a uterine perforation measuring 2 × 2 cm was found on the right fundal side of the uterus (**Figure 2**). Emergency total abdominal hysterectomy was performed. The patient underwent chemotherapy postoperatively and is currently on routine surveillance by β-hCG monitoring.

Discussion

Because every normal pregnancy includes a physiological invasion phase during placentation, myometrial invasion likely occurs in the majority of GTDs. However, it might be challenging to determine whether an aberrant myometrial invasion exists using pelvic ultrasound or suction curettage.[3] Our patient underwent a curettage procedure 3 months before admission. She was scheduled to undergo chemotherapy. However, due to poor patient compliance, chemotherapy was postponed for 3 months, and she developed uterine perforation in the emergency room.

Hydatidiform moles are the source of all invasive moles, and the majority of these moles appear within six months after the removal of the original mole. Patients with irregular vaginal bleeding and elevated serum HCG levels at this time. Approximately 15% of all invasive moles spread to the vagina or lungs. The cornerstone treatment for invasive mole is chemotherapy, whereas radiotherapy and surgery are utilized as supplemental measures. Once GTN is diagnosed, patients are grouped by FIGO Stage and World Health Organization risk score, and their stage and score dictate the initial treatment regimen to ensure the best possible outcomes with the least morbidity. Low-risk disease, defined as nonmetastatic (stage I) and low-risk metastatic GTN (stages II and III, score <7), can be managed with single-agent chemotherapy and can be considered for a second dilation and evacuation with a 40% chance of cure. High-risk disease, defined as high-risk metastatic GTN (stage IV or score ≥7), requires multi-agent chemotherapy.[7] Although the cure rate for high-risk GTN can reach 94%, the cure rate for low-risk GTN can approach 100%.[8]

Surgery is not used as an adjuvant therapy, not as a primary therapy. Hysterectomy was selected as the surgical treatment for high-risk GTN patients who do not require fertility because of its effectiveness in decreasing blood loss and reducing the complication rate. However, instead of surgery, systemic chemotherapy is usually

the first-choice treatment method for treating metastatic disease in invasive mole.[9] Our case showed an emergency situation, whereas there were acute abdominal symptoms and signs of rupture on examination. Therefore, emergency bleeding control was inevitable following hysterectomy.

The evacuation and bleeding control procedures vary depending on the situation. There was also conservative surgical treatment for uterine rupture due to GTN, such as localized resection and uterine reconstruction. Because these tumors are notoriously vascular, uterine resection is associated with a high risk of blood loss. In addition, only a few case reports have been published in the literature that have documented the effective use of resection and repair for cases of uterine rupture due to GTN.[10] Laparoscopic management of uterine perforation in hemodynamically stable patients is feasible. It also has the advantage of shorter hospital stay and earlier return to normal activities, and more advantages include laparoscopic surgery.[8] In our case, laparotomy was performed not only because of the worsening condition of the patient but also because of the size of the uterus. Based on the literature, it was stated that for a uterus size of >12 weeks of pregnancy, the best hysterectomy approach is by laparotomy.[11]

Bilateral salpingo-oophorectomy was not performed because the possibility of metastasis to the ovary is extremely low. This risk is even lower than that of nongestational primary ovarian choriocarcinoma, whose incidence is 1 in 4,000,000. Nongestational ovarian choriocarcinoma remains an important differential diagnosis. Omentectomy was not performed because metastases to the omentum are very rare in cases of invasive mole.[12] A patient with GTN had an enlarged uterus with a heterogeneous mass composed of multiple large vessels.[13] Therefore, the retroperitoneal space was not opened to minimize bleeding during the procedure. The clamp was placed as close as possible to the lateral side of the uterus to prevent ureteral injury. The primary goal of the surgery was to control hemorrhage effectively. Consequently, total abdominal hysterectomy was performed. Conservative management could not be achieved because of massive intra-abdominal blood loss. Hence, the life-saving method was selected to achieve a better outcome.

Conclusion

Due to concomitant hemorrhagic shock, uterine rupture during GTD is a rare presentation connected to an invasive form of GTD. In most cases, a hysterectomy is required for treatment. After surgery, chemotherapy is frequently necessary, but the overall prognosis is favorable. Regardless of the efficacy of chemotherapy in GTD, abdominal hysterectomy proved to be beneficial for treating uterine perforation in an emergency setting.

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