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Case Report

Two cesarean deliveries after hemi-hysterectomy due to gestational trophoblastic neoplasia

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ABSTRACT

Objective: Although uterine didelphys per se is not associated with an impaired ability to conceive, the association between uterine anomalies and gestational trophoblastic neoplasia (GTN) remains unclear. The management of chemotherapy-resistant GTN in women with uterine didelphys raises a new issue regarding whether to perform a hemi-hysterectomy.

Case report: A 23-year-old, gravida 1, para 0 Japanese woman was referred with a failed intermittent cervical dilatation for hematometra. Four years previously, she developed a GTN Stage III, score 5. As two cycles of chemotherapy with methotrexate (MTX) and one cycle of EMA-CO (etoposide, MTX, actinomycin D, cyclophosphamide and vincristine) did not result in remission, we performed an abdominal hemi-hysterectomy. After a canalization procedure and cervicoplasty were performed, the patient conceived naturally and prematurely delivered by cesarean section twice.

Conclusion: A hemi-hysterectomy should be considered for fertility preservation when GTN develops on either side of a didelphic uterus and adjuvant chemotherapy does not result in remission.

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Introduction

Uterine didelphys occurs as failure of fusion of the bilateral müllerian ducts, consequently producing duplication of the reproductive structures. It is seen in approx. 8% of all types of uterine anomalies [1]. In 15%–20% of women with uterine didelphys, unilateral anomalies are also present, such as an obstructed hemivagina and ipsilateral renal agenesis (OHVIRA syndrome) [2], Wunderlich syndrome or Herlyn-Werner syndrome. A septated vagina also occurs in 75% of women with uterine didelphys [3] and may cause difficulty with sexual intercourse or vaginal delivery.

This type of anomaly (i.e., uterine didelphys, sometimes called 'double uterus') per se is not associated with an impaired ability to conceive. When the proper management — including resection of the vaginal septum — is offered, women with uterine didelphys often have good reproductive outcomes [4]. However, little has been reported on the relationship between uterine anomalies and gestational trophoblastic disease (GTD), which could be the precursor of gestational trophoblastic neoplasia (GTN). The management of chemotherapy-resistant GTN in women with uterine didelphys raises the new issue of whether a hemi-hysterectomy should be performed.

Originally, hemi-hysterectomies were performed for women with a unicornuate uterus with a rudimentary horn (i.e., a non-communicating hemi-uterus) to alleviate dysmenorrhea, to prevent an intracornual pregnancy, and to possibly prevent endometriosis [5]. Although a hemi-hysterectomy has been performed for a small number of women with uterine didelphys and a complete bicornuate uterus who had two uterine corpi, two endometrial cavities, and two uterine cervices, the reproductive outcome of this procedure remains unclear.

We present the case of a young woman with uterine didelphys who presented a molar pregnancy with invasive and metastatic GTN, and we report her therapeutic course including the hemi-hysterectomy and reproductive outcome.

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Case presentation

A 23-year-old, gravida 1, para 0 Japanese woman was referred to Oita University Hospital with a failed intermittent cervical dilatation for cervical occlusion and hematometra which had resulted in the gradual development of hypomenorrhea and dysmenorrhea over a 1-year period. She had a noteworthy medical history.

Four years previously, she conceived naturally, and uterine didelphys with a longitudinal vaginal septum first diagnosed at Oita Red Cross Hospital. She underwent uterine evacuations for a presumed molar pregnancy in the left hemiuterus, which was diagnosed as a partial mole based on the microscopic findings of the focal hydropic change of chorionic villi. However, at her follow-up visit, she displayed nausea, and her urinary human chorionic gonadotropin (hCG) level was identified as increased to 3,276,800 mIU/mL. A hypervascular tumor in her left hemiuterus measuring 50 mm in dia. (Fig. 1A) and multiple nodules in bilateral lungs (Fig. 1B) were shown by contract-enhanced computed tomography (CECT). Dr. E. Hori (Oita Red Cross Hospital) diagnosed gestational trophoblastic neoplasia (GTN) Stage III, score 5. As two cycles of chemotherapy with methotrexate (MTX) with folinic acid did not result in remission, an EMA-CO regimen of etoposide, MTX, actinomycin D, cyclophosphamide and vincristine was initiated. After the 1st cycle of EMA-CO, her urinary hCG value rose and multiple pulmonary nodules increased in size, contrary to our expectations. She underwent abdominal hemi-hysterectomy and a resection of vaginal septum. Hydropic chorionic villi, some of which were invading in the myometrium with trophoblastic proliferation suggesting an invasive mole, were shown by a microscopic examination (Fig. 1C). A diploid diandric genome suggesting a complete hydatidiform mole was confirmed by a DNA genotyping test. After five adjuvant cycles of EMA-CO, complete remission was achieved. During the 2-year period of post-treatment severance, the patient had a regular menstrual cycle and was free from relapse.

On her initial visit to our unit (Oita University Hospital), a speculum examination did not show any menstrual blood in her vagina, but there was menstrual blood in the uterine cavity (hematometra). The uterine cervix with the residual side was unclear. A transvaginal sonography showed an extremely right-deviated uterus measuring 50 mm in dia. Magnetic resonance imaging revealed a retained right uterus with a hypoplastic cervix and right adnexa (Fig. 2A), and CECT demonstrated normal bilateral kidneys (Fig. 2B). We concluded that her primary disease was uterine didelphys with a hypoplastic right hemi-uterus at the cervix, bicollis, with vaginal septum (1988 American Fertility Society classification type III). We performed a canalization procedure with cervical dilators and a cervicoplasty under general anesthesia. Despite ultrasound guidance, we caused an intraoperative bladder injury. After 10 days of postoperative cervical canal canalization with a 14-Fr soft catheter, the patient was discharged uneventfully.

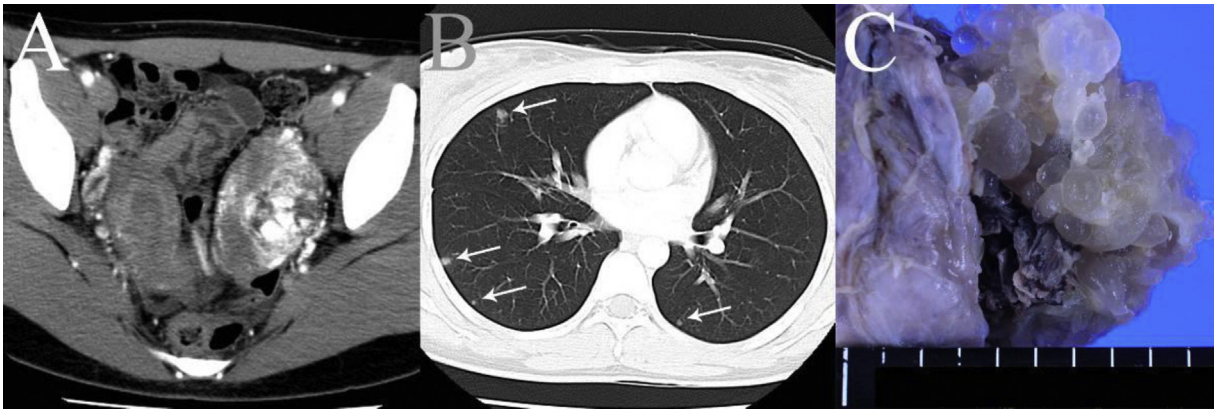

Fig. 1. A: Uterine didelphys and a hypervascular tumor occupying the intrauterine cavity of the left hemiuterus. **B:** Multiple and bilateral pulmonary metastases of gestational trophoblastic neoplasia (GTN) by contract-enhanced computed tomography. **C:** The resected specimen showed an aggregate of branching edematous villi.

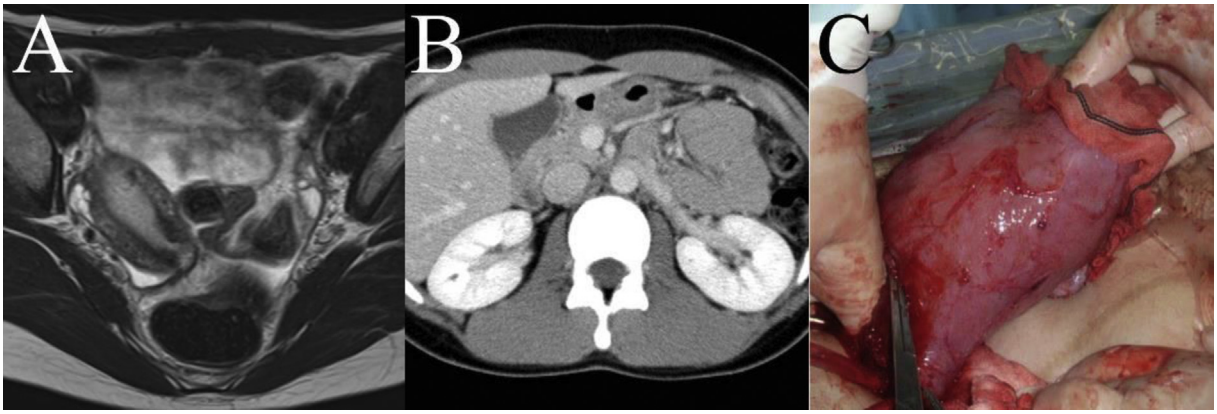

Fig. 2. A: The retained right uterus with a hypoplastic cervix. **B:** Bilateral kidneys were present without abnormal findings. **C:** Neither adnexa nor a suspensory ligament of the ovary was noted on the left side of the uterus at the time point of cesarean section.

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