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Septic shock in a woman with a hydatidiform mole: A case report

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ABSTRACT

Hydatidiform moles can be fatal because of the risk of massive bleeding or thyroid storm; however, they rarely occur concomitantly with sepsis. We present herein the case of a woman with a hydatidiform mole with septic shock. A 30-year-old multiparous woman with Basedow's disease presented with fever, amenorrhea, and vaginal bleeding. Transvaginal ultrasonography revealed an enlarged uterus with an intrauterine vesicular mass (74.3 × 93.0 mm). Human chorionic gonadotropin level was 994,000 mIU/mL. C-reactive protein was elevated, and blood cultures were positive (gram-negative rods), indicating infection. After administering antibiotics (tazobactam and piperacillin), blood pressure suddenly decreased (69/45 mmHg), requiring stabilization with noradrenaline and albumin. The uterine contents were naturally expelled, followed by dilatation and curettage after her vital signs and general condition gradually improved. The pathological diagnosis was a complete hydatidiform mole. Culture of the intrauterine contents revealed *Escherichia coli*, leading to the potentially fatal diagnosis of septic shock associated with a hydatidiform mole.

1. Introduction

Hydatidiform mole is a placental pathology of androgenetic origin, wherein placental villi undergo abnormal hyperproliferation and hydropic degeneration. Hydatidiform moles can be fatal (especially in those with advanced gestational age) because of the occurrence of massive bleeding or thyroid storm [1,2]. Meanwhile, other potentially lethal conditions that can occur in women with hydatidiform moles should be elucidated. The association between hydatidiform moles and sepsis has been rarely reported. Herein, we present a case of septic shock associated with a hydatidiform mole in a woman with a history of thyroid disease.

2. Case Presentation

A 30-year-old multiparous woman (two uneventful spontaneous deliveries and one spontaneous abortion) with a history of Basedow's disease (stable condition with propylthiouracil 300 mg/day) presented to a general internal medicine doctor because of fever (38.5 °C) lasting 3 days. The polymerase chain reaction test for coronavirus disease 2019 (COVID-19) was negative. Her vital signs were as follows: pulse rate, 82 beats per minute (bpm); blood pressure, 111/72 mmHg; and respiratory rate, 16 breaths per minute. Blood samples were collected, and blood cultures were performed. The laboratory data showed a white blood cell (WBC) count of 8.9×10^3/μL, hemoglobin level of 10.5 g/dL, platelet count of 210×10^3/μL, C-reactive protein (CRP) level of 3.0 mg/dL, thyroid-stimulating hormone level < 0.1 μIU/mL (reference level 0.35–4.94 μIU/mL), triiodothyronine level of 3.65 pg/mL (reference level 1.88–3.18 pg/mL), and thyroxine level of 1.84 ng/dL (reference level 0.70–1.48 ng/dL). On physical examination, she had tenderness in the lower quadrant of the abdomen and enlargement of the thyroid gland. Admission and further evaluation were suggested; however, the patient declined this for personal reasons.

The next day, she contacted the general medicine doctor again. Her vital signs were as follows: body temperature, 36.6 °C; pulse rate, 109 bpm; blood pressure, 104/63 mmHg; and respiratory rate, 16 breaths per minute. Blood cultures (midterm report) were positive for gram-negative rods, indicating infection. She had tenderness in the lower quadrant of the abdomen. Since a thorough interview revealed a history of amenorrhea for 10 weeks and vaginal bleeding for 4 weeks, a pregnancy test was performed, which was positive, necessitating a gynecological consultation.

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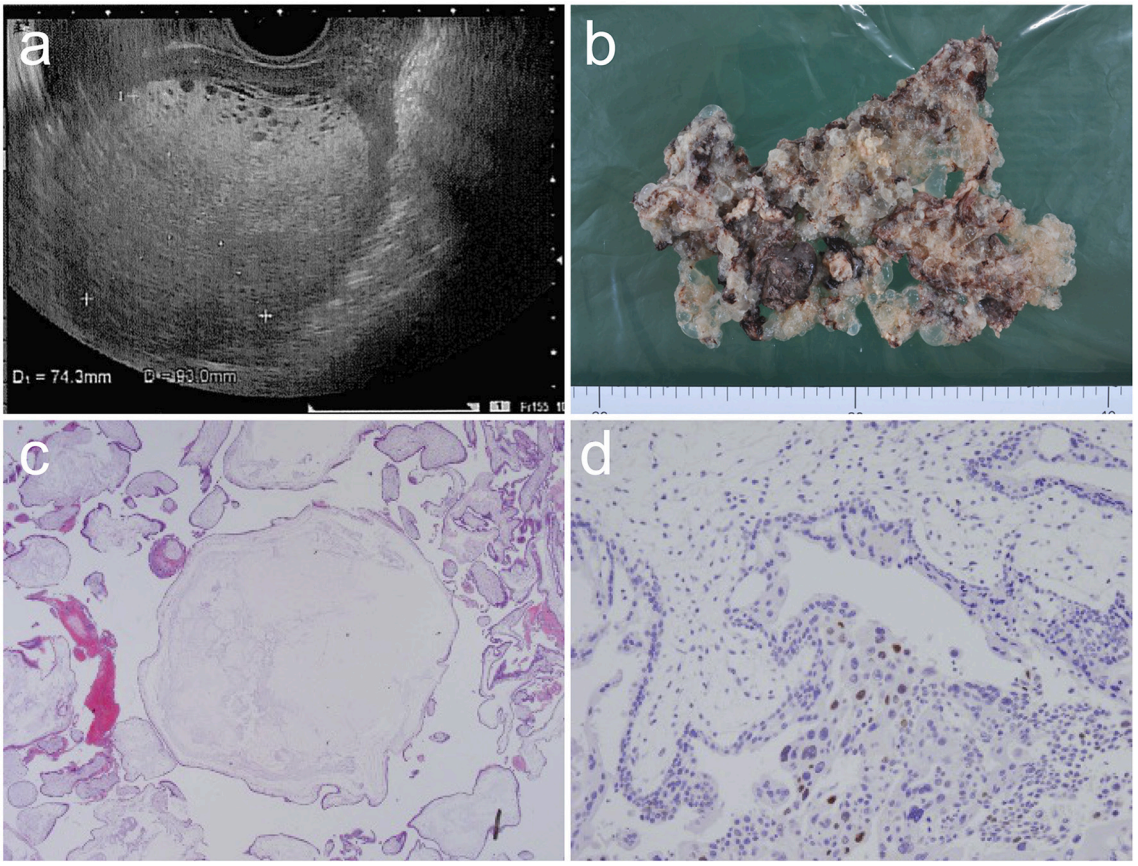

Fig. 1. a–d. Transvaginal ultrasonography at the initial visit shows an enlarged uterus with an intrauterine mass (74.3 × 93.0 mm), with a vesicular pattern (a). A substantial amount of cystic uterine contents (approximately 500 g) was naturally expelled after the patient was treated for septic shock (b). Pathologically, the complete hydatidiform mole shows abnormal hyperproliferation of the placental villi and hydropic degeneration associated with the absence of an embryo (c). On immunohistological examination, the complete hydatidiform mole was p57-negative (d).

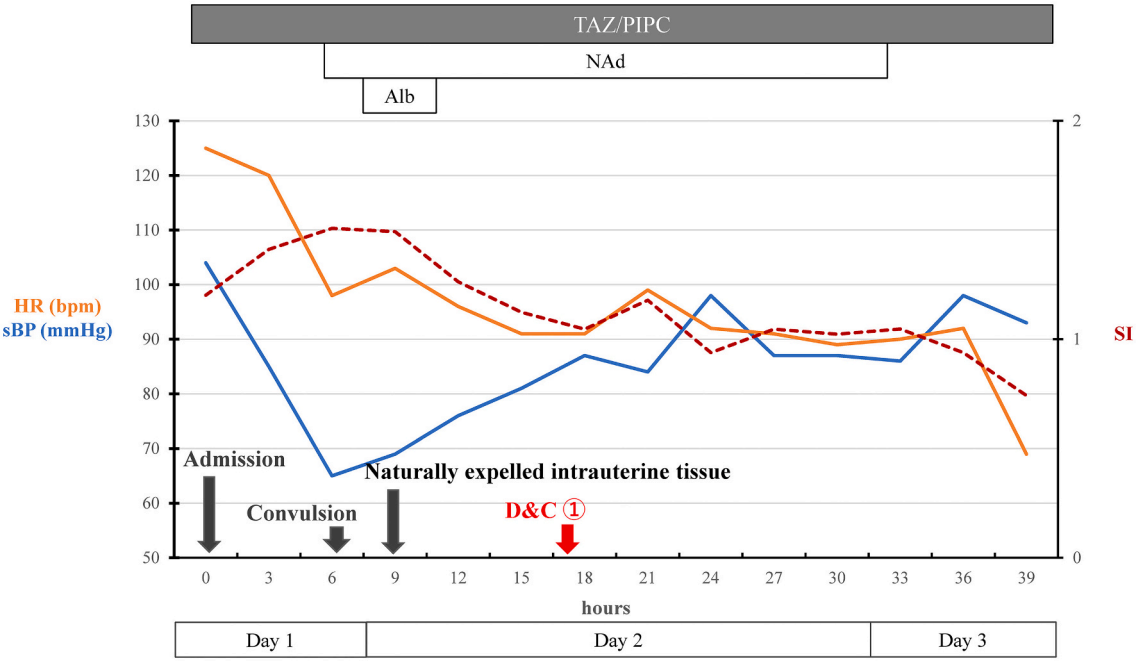

Fig. 2. Clinical course during hospitalization.
Abbreviations: Alb, albumin; bpm, beat per minute, D&C, dilation and curettage, HR, heart rate, NAd, noradrenaline, SBP, systolic blood pressure, SI, shock index, TAZ/PIPC, tazobactam and piperacillin.

Transvaginal ultrasonography revealed an enlarged uterus with an intrauterine mass (74.3 × 93.0 mm), with a vesicular pattern (Fig. 1a), normal bilateral ovaries, and no ascites. Laboratory tests revealed a human chorionic gonadotropin (hCG) level of 994,000 mIU/mL, WBC count of 12.1 × 10^3/μL, hemoglobin level of 10.2 g/dL, platelet count of 133 × 10^3/μL, and CRP of 15.1 mg/dL. Computed tomography demonstrated no other potential sites of infection or hydatidiform mole metastases in the body. She was admitted to the gynecological unit and administered antibiotics (tazobactam and piperacillin 4.5 g every 8 h). Additionally, dilation and curettage (D&C) was planned.

However, her blood pressure suddenly plummeted to 69/45 mmHg with tachycardia (pulse rate of 115 bpm) 6 h after the admission, followed by a brief episode of convulsions with rapid improvement in her consciousness. Septic shock was considered and the patient was administered noradrenaline (0.1 μg/kg/min), albumin (12.5 mg/250 ml), and appropriate fluid resuscitation to maintain blood pressure. During the preparation for D&C by cervical dilatation and vital signs stabilization, a large amount of cystic uterine tissue (approximately 500 g) was naturally expelled approximately 3 h after decreased blood pressure had been detected (Fig. 1b). Her vital signs remained unstable; however, abdominal pain and vaginal bleeding improved 12 h after the onset, and the vital signs gradually stabilized (pulse rate 89 bpm and blood pressure 91/59 mmHg). The total amount of genital bleeding after hospitalization was less than 100 mL, which excluded hypovolemic shock due to massive genital bleeding. D&C was performed twice (days 2 and 10, using pentazocine 15 mg and propofol 100 mg) following stabilization of the vital signs. The expelled uterine contents and the tissue obtained in the first D&C were pathologically examined, and a diagnosis of a complete hydatidiform mole was established based on enlarged hydropic villi (Fig. 1c) and negative p57 immunostaining (Fig. 1d). The second D&C yielded the remaining necrotic chronic degenerative tissue. Blood culture and culture of the intrauterine contents revealed *E. coli* infection, leading to the diagnosis of septic shock associated with a hydatidiform mole.

Noradrenaline was administered for 24 h until the patient's condition stabilized (Fig. 2). Antibiotics were administered for 8 days, until an improvement in the laboratory parameters was confirmed. Her thyroid function and enlargement of the thyroid gland normalized on day 10. The patient was discharged 11 days after hospitalization. Ten days after hospital discharge, the hCG level had decreased to 390 mIU/mL. However, the patient refused a follow-up hospital visit based on her judgment. Written informed consent was obtained from the patient for the publication of this report.

3. Discussion and Conclusions

The present case demonstrates the possibility of septic shock occurring with a hydatidiform mole, which can be a life-threatening condition. In addition to massive bleeding and thyroid storm, infection can be a lethal complication in women with hydatidiform moles.

The association between hydatidiform moles and sepsis has been rarely reported. Adams et al. reported the case of a 49-year-old multiparous woman with a hydatidiform mole at 20 weeks of gestation who developed sepsis due to an intrauterine *Clostridium perfringens* infection [3]. Since antibiotics were ineffective, the patient underwent hysterectomy. In the present case, sepsis associated with the hydatidiform mole was caused by *E. coli*, which is the typical bacterium responsible for endometritis [4]. Although *E. coli* sepsis can result in severe endotoxic shock [5], rapid intervention using antibiotics, natural expulsion of uterine contents, and double D&C improved the outcome, and preservation of the uterus was successful in this case. The possible mechanism of sepsis in hydatidiform mole is based on endometritis, which could be caused by cervical opening with continuous genital bleeding and increasing intrauterine pressure by intrauterine mass with advancing gestational age. In the present case, a large hydatidiform mole obstructing the cervix and increasing the intrauterine pressure may have led to sepsis. Meanwhile, septic shock might not have been essentially caused by a hydatidiform mole; rather, it might have been caused by the possibility of hydatidiform mole as a risk factor for exacerbation of intrauterine infection with continuous genital bleeding [6]. Therefore, the detailed etiology of sepsis in a hydatidiform mole case should be clarified through further studies.

In addition to sepsis, several lethal conditions can occur in patients with a hydatidiform mole. A literature review identified numerous case reports of hydatidiform moles with massive bleeding or hyperthyroidism, which can be fatal. D&C of hydatidiform mole can cause uterine perforation, which can result in massive bleeding [1]. Higher levels of hCG, which has thyrotropic activity, can cause thyroid crisis and multiorgan failure [2]. Specifically, hydatidiform mole with advanced gestational age is associated with an increased risk of active bleeding and increased stimulation of the thyroid [7] and requires urgent intervention compared to that with earlier gestation. Thus, advanced gestational age in patients with hydatidiform mole would lead to higher risks of lethal conditions, including larger tumors, resulting in an increased risk of septic conditions.

Interestingly, there was a significant two-fold increase in the incidence of molar pregnancy during the COVID-19 pandemic compared with that in the previous 10 years [8]. This could be because visiting the physician in the later stage of an abnormal pregnancy can clearly distinguish abortion from molar pregnancy by specific sonographic findings and higher serum hCG levels, which could lead to an increase in the number of uterine evacuations for suspected molar pregnancies [8]. This phenomenon should reflect the fact that the COVID-19 pandemic would lead to a delayed visit to the physician. Thus, we should encourage patients to visit the hospital early despite the pandemic when experiencing abdominal pain, large amounts of genital bleeding, and fever because a hydatidiform mole can become a lethal condition over time.

In conclusion, a hydatidiform mole can cause septic shock, especially in those at advanced gestational age. We should consider the possibility of septic shock in patients with hydatidiform moles in addition to the risk of massive bleeding and endocrine disorders, and rapid intervention with antibiotics and D&C should be provided.

Contributors

Yuki Yoshimoto contributed to patient management and data collection, and drafted the manuscript.

Tsuyoshi Murata contributed to patient management and editing of the manuscript.

Moeko Machida contributed to patient management and editing of the manuscript.

Yoshihiro Nozawa contributed to histopathological analysis and editing of the manuscript.

Soichi Nakamura contributed to patient management and editing of the manuscript.

Ryuji Yamauchi contributed to patient management and editing of the manuscript.

All authors approved the final article to be submitted.

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Patient consent

Written informed consent was obtained from the patient for the publication of this case report.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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