

CASE REPORT

Successful treatment of primary non-gestational choriocarcinoma of the uterine cervix in a postmenopausal woman

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Abstract

Background: Primary non-gestational choriocarcinoma of the uterine cervix is an exceedingly rare malignancy, particularly in postmenopausal women, and is associated with a poor prognosis. Owing to its rarity, standardized treatment guidelines for this disease have not been established. **Case:** A 52-year-old woman presented with vaginal bleeding and was found to have a 4.6-cm cervical mass with markedly elevated serum β -human chorionic gonadotropin (β -hCG). Imaging demonstrated disease confined to the uterine cervix, consistent with the International Federation of Gynecology and Obstetrics (FIGO) stage IB2 (revised IB3). The patient underwent total hysterectomy with bilateral salpingo-oophorectomy, and histopathologic examination confirmed primary non-gestational choriocarcinoma. Adjuvant chemotherapy with bleomycin, etoposide, and cisplatin (BEP) was administered for three cycles, resulting in normalization of serum β -hCG. At eight years of follow-up, the patient remains disease-free. **Conclusion:** We report the successful management of primary non-gestational choriocarcinoma of the uterine cervix in a postmenopausal woman using hysterectomy followed by adjuvant BEP chemotherapy, consistent with therapeutic principles for malignant germ cell tumors.

Keywords

Postmenopausal woman; Uterine cervix; Primary non-gestational choriocarcinoma; Treatment

1. Introduction

Primary non-gestational choriocarcinoma of the uterine cervix is an exceptionally rare malignancy and is associated with a poorer prognosis compared with gestational choriocarcinoma of the genital tract [1]. Owing to its rarity and overlapping clinical features, the disease may be easily misdiagnosed as cervical pregnancy or other cervical malignancies [1–3]. Because of diagnostic challenges, reported cases have often been managed with concurrent chemoradiation, as is standard for common cervical malignancies, or with systemic chemotherapy regimens based on methotrexate and/or actinomycin D, following the therapeutic principles of gestational trophoblastic disease [1, 3]. However, primary non-gestational choriocarcinoma of the uterine cervix is thought to arise from germ cells and, therefore, exhibits biological behavior similar to that of malignant germ cell tumors [3, 4].

Here, we present a case of primary non-gestational choriocarcinoma confined to the uterine cervix in a postmenopausal woman, managed according to treatment strategies established for gynecological malignant germ cell tumors.

2. Case report

A 52-year-old woman (gravida 3, para 3) was admitted to the Department of Gynecologic Oncology at Chonnam National University Hwasun Hospital in July 2017, with a chief complaint of vaginal bleeding for approximately two months. Her medical history was notable only for hypertension. She had no prior surgeries and no other significant comorbidities. She had experienced amenorrhea for two years and was presumed to be in the menopausal transition, although menopausal status had not been biochemically confirmed. She had three full-term spontaneous vaginal deliveries, the first at age 26 and the last at age 36.

On admission, her vital signs were stable, with a blood pressure of 140/70 mmHg, heart rate of 92 beats/min, respiratory rate of 20 breaths/min, and body temperature of 36.3 °C. Pelvic examination revealed a dark hemorrhagic tumor measuring approximately 5 cm located on the anterior lip of the uterine cervix (Fig. 1). Laboratory evaluation showed a hemoglobin level of 11.5 g/dL (reference range: ≥ 12.0 g/dL), with all other parameters within normal limits. A urine pregnancy test was positive, prompting further evaluation with a serum β -human chorionic gonadotropin (β -hCG) assay, which revealed

a markedly elevated level of 12,900 mU/mL (reference range: <5 mU/mL). Other serum cervical tumor markers, including squamous cell carcinoma antigen (SCC-Ag), 0.7 ng/mL; cancer antigen 125 (CA-125), 25 U/mL; and carcinoembryonic antigen (CEA), 1.2 ng/mL, were within normal limits. The patient underwent a uterine cervical biopsy, and the specimen was interpreted as suspicious for choriocarcinoma.

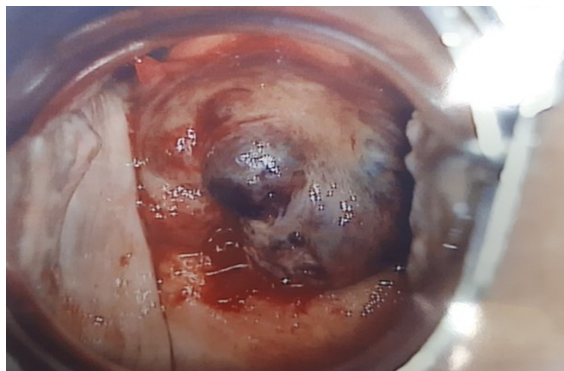


FIGURE 1. Pelvic examination revealed a dark, hemorrhagic mass measuring approximately 5 cm on the anterior lip of the uterine cervix.

Chest radiography, pelvic magnetic resonance imaging (MRI), abdominal and chest computed tomography (CT), and brain MRI were performed. Pelvic MRI revealed a 4.6-cm trophoblastic tumor confined to the anterior lip of the uterine cervix (Fig. 2). All other imaging studies showed no evidence of involvement of other organs. The disease was staged as IB2 according to the International Federation of Gynecology and Obstetrics (FIGO) system, corresponding to stage IB3 in the revised FIGO classification [5].

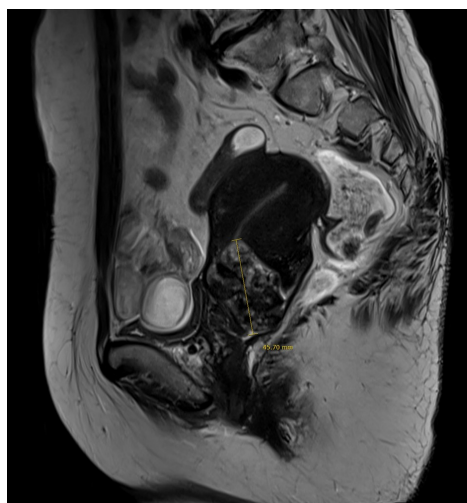


FIGURE 2. Pelvic magnetic resonance imaging (MRI) demonstrated a heterogeneously enhancing, ill-defined mass measuring $3.7 \times 3.4 \times 4.6$ cm located at the anterior lip of the uterine cervix. On T2-weighted imaging, the lesion exhibited heterogeneous signal intensity with areas of intratumoral hemorrhage. Diffusion restriction was not prominent. The mass appeared to invade both cervical stromal regions.

The patient underwent laparotomic total hysterectomy with bilateral salpingo-oophorectomy. Gross examination revealed a 5-cm dark, hemorrhagic, necrotic mass with full-thickness stromal invasion of the uterine cervix (Fig. 3). The postoperative course was uneventful, and the patient was discharged on the fifth postoperative day. At that time, the serum β -hCG level had decreased to 6270 mU/mL. Histopathological examination confirmed choriocarcinoma. Microscopic evaluation demonstrated an atypical admixture of cytotrophoblasts and syncytiotrophoblasts, and immunohistochemistry revealed positive staining for β -hCG (Fig. 4). The final pathology established a diagnosis of FIGO stage IB2 primary non-gestational choriocarcinoma of the uterine cervix (corresponding to stage IB3 under the revised FIGO staging system) [5].

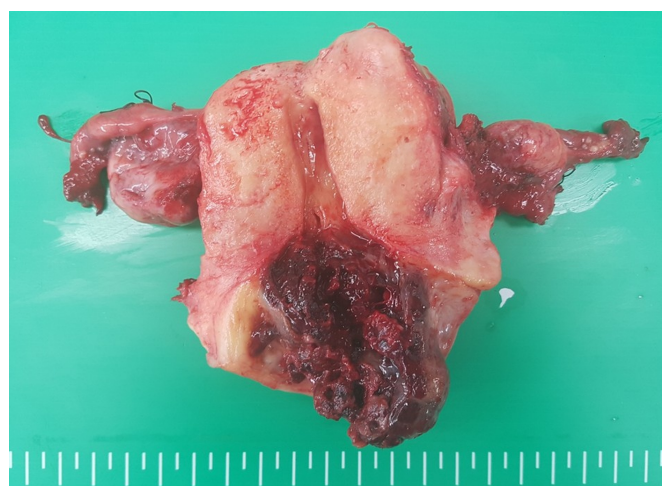


FIGURE 3. Gross examination of the uterine cervical tumor revealed a dark, hemorrhagic, necrotic mass measuring approximately 5 cm in diameter. A bisected view of the excised uterus showed full-thickness stromal invasion of the cervix. Both ovaries and fallopian tubes appeared intact, with no gross abnormalities.

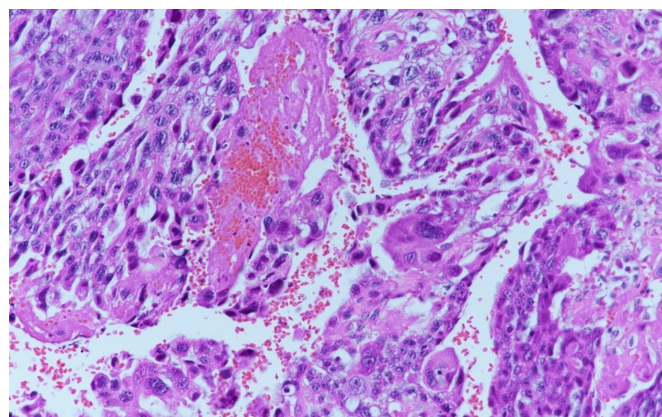


FIGURE 4. Histopathological examination demonstrated an atypical admixture of cytotrophoblasts and syncytiotrophoblasts (hematoxylin and eosin, $\times 100$). Immunohistochemical staining showed positive expression of β -human chorionic gonadotropin (β -hCG).

Following multidisciplinary tumor board discussion, and in accordance with therapeutic principles for gynecological malignant germ cell tumors, systemic chemotherapy with bleomycin, etoposide, and cisplatin (BEP) was initiated one month after surgery. The patient received three cycles of BEP chemotherapy, consisting of bleomycin (15 units/m² intravenously on days 1, 8, and 15), etoposide (100 mg/m² intravenously on days 1–5), and cisplatin (20 mg/m² intravenously on days 1–5), administered every 21 days. Table 1 shows treatments administered and serial serum β -hCG levels.

Following systemic chemotherapy, the patient's serum β -hCG level decreased to ≤ 1 mU/mL. Follow-up abdominal and chest CT performed six months later revealed no abnormalities, and serum β -hCG levels remained within normal limits. As of July 2025, at eight years of follow-up, the patient remains well and free of disease recurrence.

3. Discussion

Primary non-gestational choriocarcinoma of the uterine cervix is extremely rare and poses substantial diagnostic challenges [1]. Because of its rarity, it is often misdiagnosed as cervical pregnancy, missed abortion, necrotic cervical polyp, cervical myoma, or other forms of cervical carcinoma [2]. This diagnostic difficulty frequently results in delayed diagnosis and inappropriate management. In particular, when non-gestational choriocarcinoma is not distinguished from its gestational counterpart, patients may fail to receive optimal therapy and consequently experience poor outcomes. Compared with gestational choriocarcinomas, non-gestational tumors are typically more aggressive, exhibit a higher metastatic potential, and demonstrate reduced responsiveness to chemotherapy [1, 6, 7].

The pathogenesis of non-gestational choriocarcinoma has been hypothesized to involve aberrant germ cells that fail to complete their migration to the gonads [8]. Alternatively, primary cervical disease may arise through metaplastic transformation of the cervical epithelium, dedifferentiation of native cervical cells into choriocarcinoma, or metastasis to the cervix from a primary tumor of the uterine corpus [9, 10]. Definitive diagnosis relies on histopathological evaluation. Microscopically, these tumors characteristically demonstrate a malignant admixture of cytotrophoblastic and syncytiotrophoblastic cells [11, 12]. Immunohistochemical staining serves as an essential diagnostic adjunct, with tumor cell positivity for β -hCG and human placental lactogen (hPL) supporting the diagnosis of

non-gestational choriocarcinoma [11, 12].

Surgical debulking followed by adjuvant chemotherapy is generally regarded as the standard treatment for non-gestational choriocarcinoma of the genital tract. The treatment of primary non-gestational choriocarcinoma of the uterine cervix does not seem to deviate from surgery and followed systemic chemotherapy. In one study, a patient with the tumor was treated with hysterectomy followed by Etoposide, Methotrexate, Actinomycin D, Cyclophosphamide, and Vincristine (EMA/CO) chemotherapy, analogous to treatment strategies for gestational choriocarcinoma [1, 3, 7, 10, 12, 13]. Chemoradiation following hysterectomy—an approach aligned with management of common epithelial cervical cancers—has also been reported [1, 3]. However, such strategies may not be appropriate for non-gestational choriocarcinoma, given its distinct biology. Because these tumors may arise through dedifferentiation from another histologic type, their responsiveness to chemotherapy and radiotherapy differs from that of both gestational choriocarcinomas and conventional cervical cancers [14].

Table 2 (Ref. [3, 4, 9, 10]) shows primary non-gestational choriocarcinoma of the uterine cervix in articles published in the English literature over the past twenty years. The management of non-gestational choriocarcinoma of the uterine cervix should be aligned with the therapeutic principles applied to non-gestational choriocarcinomas arising in other reproductive organs. Numerous studies have demonstrated that malignant germ cell tumors respond favorably to bleomycin, etoposide, and cisplatin (BEP) chemotherapy [15]. Accordingly, the treatment of primary non-gestational choriocarcinoma of the uterine cervix should follow the same strategy. A previously reported case of cervical choriocarcinoma also described the successful use of BEP chemotherapy [6]. Based on this evidence, and after comprehensive multidisciplinary discussion at the tumor board, adjuvant BEP chemotherapy was selected for this patient following surgical resection. Consequently, she has remained disease-free and in good health for eight years after treatment.

As with other gynecologic malignancies, the stage of disease at diagnosis represents an important prognostic factor [3, 4]. Most reported cases have involved patients presenting with advanced-stage disease (Table 2). Clinical manifestations of choriocarcinoma in postmenopausal women are often nonspecific. Although postmenopausal bleeding is the most common presenting symptom, some cases have been identified in the context of acute surgical abdomen [11–14]. In certain in-

TABLE 1. Treatments administered and serial serum β -human chorionic gonadotropin (β -hCG) levels.

Date	Treatment	β -hCG (mU/mL)
16 August 2017	Total hysterectomy with bilateral salpingo-oophorectomy	12,900
31 August 2017		55
15 September 2017	1st bleomycin, etoposide, and cisplatin	7
13 October 2017	2nd bleomycin, etoposide, and cisplatin	<1
10 November 2017	3rd bleomycin, etoposide, and cisplatin	1
Since 30 November 2017		≤ 1

Reference range, β -hCG <5 mU/mL.

TABLE 2. Primary non-gestational choriocarcinoma of the uterine cervix in articles published in the English literature over the past twenty years.

Articles	Age (yr)	Tumor size (cm)	FIGO stage	Initial β -hCG (mU/mL)	Treatment	Outcome
Hwang <i>et al.</i> [10]	45	8	IVB	11,924	Paclitaxel + carboplatin $\times 3$ RH + BSO + PLND (massive bleeding) Adjuvant EMA/CO $\times 6$ Whole brain radiotherapy	Death of disease progression
Longo <i>et al.</i> [4]	54	-	IVB	29,976	RH and adjuvant BEP $\times 4$ Relapse 2 months later Cisplatin + vinblastine + ifosfamide $\times 2$ Whole brain radiotherapy	Death of disease progression
Chumworathayi <i>et al.</i> [9]	47	8	IB3	8	Actinomycin D $\times 5$ and RH + PLND	Alive
Maesta <i>et al.</i> [3]	32	11	IVB	451,649	EMA/CO $\times 5$ EMA/EP $\times 4$ Paclitaxel + cisplatin + etoposide $\times 2$ Paclitaxel + carboplatin $\times 1$ Gemcitabine $\times 1$ Taxotere + 5FU $\times 1$	Death of disease progression

FIGO, the International Federation of Gynecology and Obstetrics; β -hCG, β -human chorionic gonadotropin; RH, radical hysterectomy; BSO, bilateral salpingo-oophorectomy; PLND, pelvic lymph node dissection; EMA/CO, etoposide, methotrexate, actinomycin D, cyclophosphamide and vincristine; BEP, bleomycin, etoposide and cisplatin; EMA/EP, etoposide, methotrexate, actinomycin D, etoposide and cisplatin; 5FU, 5-Fluorouracil.

stances, clinical presentation may be dominated by metastatic disease, most frequently involving the lungs, but also the brain, liver, digestive tract, or urinary system [12]. Measurement of serum β -hCG is valuable for both diagnosis and monitoring, although it is not routinely performed in postmenopausal women [3, 12].

To date, standardized treatment guidelines for primary non-gestational choriocarcinoma of the uterine cervix have not been established, largely due to the extreme rarity of the disease. This case contributes important evidence supporting the use of adjuvant BEP chemotherapy following hysterectomy as an effective therapeutic approach for this rare entity.

4. Conclusion

Primary non-gestational choriocarcinoma of the uterine cervix is an exceptionally rare malignancy, and defining an optimal treatment strategy remains a significant challenge. As a result, prior reports have frequently adopted therapeutic approaches used for gestational choriocarcinoma or for common epithelial cervical cancers. Although the present case involved an early-stage tumor, to the best of our knowledge, it is the first to demonstrate a favorable long-term outcome following hysterectomy with adjuvant bleomycin, etoposide, and cisplatin (BEP) chemotherapy in primary non-gestational choriocarcinoma of the uterine cervix. This case report has inherent limitations, including its single-patient design due to limited generalizability. Additional case reports and larger studies will be essential to establish evidence-based management guidelines for this rare entity.

AVAILABILITY OF DATA AND MATERIALS

All data supporting the findings of this case report are included in the article and available from the corresponding author upon reasonable request.

AUTHOR CONTRIBUTIONS

UJ, EH, WDK and SMK—designed the research study; performed the research and analyzed the data. WDK and SMK—provided guidance and advice on data analysis. UJ and EH—wrote the manuscript. All authors have read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The protocol was approved by the Institutional Review Board of Chonnam National University Hwasun Hospital (No. CNUHH-2025-081). Informed consent for participation and publication was obtained from the patient.

ACKNOWLEDGMENT

We thank all the peer reviewers and editors for their valuable opinions and suggestions.

FUNDING

This research received no external funding.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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How to cite this article: Uchul Ju, Enoch Hong, Woo Dae Kang, Seok Mo Kim. Successful treatment of primary non-gestational choriocarcinoma of the uterine cervix in a postmenopausal woman. *European Journal of Gynaecological Oncology*. 2026; 47(3): 107-111. doi: 10.22514/ejgo.2026.032.