Case Report

Successfully Conservative Treatment of Large Cervical Choriocarcinoma with Profuse Vaginal Bleeding

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Primary choriocarcinoma of the uterine cervix is a rare disease. The accurate diagnosis of such a disease is difficult to achieve because of its rarity. Furthermore, the majority of cases presented with abnormal vaginal bleeding that could be caused by other more common conditions including, threatened abortion, cervical polyp, cervical pregnancy, or cervical cancer. In the present report, the authors present a case of large cervical choriocarcinoma with life-threatening vaginal bleeding, which was initially misdiagnosed as a cervical cancer. The active cervical bleeding was successfully controlled with selective uterine arterial embolization. Remission of cervical choriocarcinoma was accomplished with combination chemotherapy without the need of hysterectomy.

Keywords: *Primary choriocarcinoma, Conservative treatment, Vaginal bleeding*

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Gestational trophoblasic disease (GTD) comprises a spectrum of abnormal proliferation of trophoblastic cells ranging from benign to malignant potential. Choriocarcinoma is a malignant trophoblastic tumor that commonly arises within the uterine corpus. On rare occasions, choriocarcinoma may occur at various sites including the uterine cervix, fallopian tube, ovary, vagina, vulva, and extragenital organs[1-8]. In the present report, the authors present a case of large cervical choriocarcinoma with life-threatening vaginal bleeding who was successfully treated with conservative management.

Case Report

A 33-year-old women (gravida 3, Para 1-0-1-1) came to the provincial hospital with the complaint of massive vaginal bleeding for 4 weeks. She irregularly used oral combined pills for contraception. Last pregnancy ended with a spontaneous abortion at 8 weeks gestation and was 8 months before. The post-abortion curettage was not performed. Last menstruation period was unknown. She had approximately 4 months of missed period before the occurrence of vaginal bleeding. She had no previous history of GTD. Pelvic examination at the provincial hospital revealed a cervical mass of 8 cm in diameter. A punch biopsy was done and subsequently reported as poorly differentiated squamous cell carcinoma. Because of her history of missed menstruation, urine pregnancy test was carried out and was positive. Vaginal packing was performed after biopsy due to massive vaginal bleeding. The patient was then referred to Chiang Mai University Hospital with the diagnosis of cervical carcinoma and early pregnancy. At Chiang Mai University Hospital, she had marked anemia with tachypnea. Pelvic examination showed a large irregular hemorrhagic mass of 8x6x5 cm at the apex of the vaginal canal with active

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bleeding. The normal cervix could not be identified. The vaginal mucosa was clinically not involved. The uterus and both adnexae were normal. Both parametrium were soft and smooth. Vaginal packing was performed again after the examination and blood transfusion was immediately provided. Chest x-ray was taken which showed multiple pulmonary nodules. Serum beta-hCG was 45,725 IU/L. The pelvic ultrasonographic examination revealed a cervical mass of 8 cm in largest diameter with high intratumoral vascularity. The uterus was normal in size and had no intrauterine content. No adnexal mass was observed. The abdominal ultrasonography and computed tomography scan of brain were unremarkable. The pathology slide review with additional sections of hCG staining showed choriocarcinoma (Fig. 3 shows the histologic findings of choriocarcinoma). According to the FIGO staging system, the diagnosis of this patient was cervical choriocarcinoma stage III; 11. However, despite the continuous vaginal packing, the massive vaginal bleeding was persistent requiring multiple blood transfusions. The intervention radiologist was consulted for selective uterine arterial embolization to induce hemostasis. The combination chemotherapy including etoposide, metrotrexate, and actionomycin-D (EMA) was administered on the first day postoperatively. Vaginal packing could be removed at 48 hours after uterine arterial embolization. The patient received 8 courses of EMA regimen with favorable response and no major adverse toxicity. Fig. 1 shows the colposcopic image of the cervix after the first course of chemotherapy and fig. 2 shows the healed cervix after completion of eight courses of chemotherapy. The serum beta-hCG level declined continuously and reached normal after six courses of chemotherapy. An additional two course were given as maintenance chemotherapy. At the completion of treatment, chest x-ray and pelvic examination were normal. Serum beta-hCG was less than 1 IU/L. The patient was well without any sign of recurrence at 33 months of the follow-up.

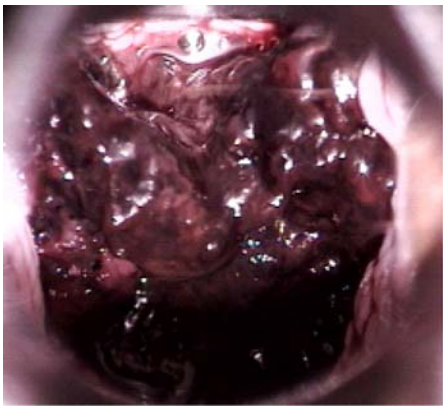

Fig. 1 Cervical choriocarcinoma: A colposcopic image of a cervix after the first course of chemotherapy (including selective uterine artery embolization). There is a large, dark exophytic mass involving the whole cervix. The patient had no vaginal bleeding

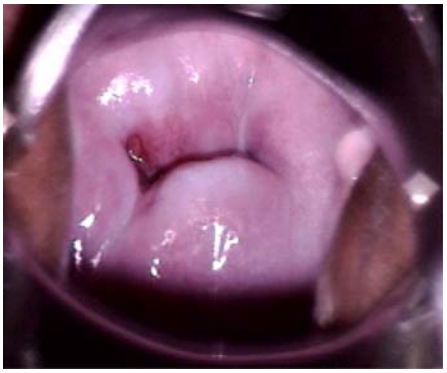

Fig. 2 The healed cervix: A colposcopic image of a cervix after completion of 8 courses of chemotherapy. The cervical contour had a great healing and becomes normal

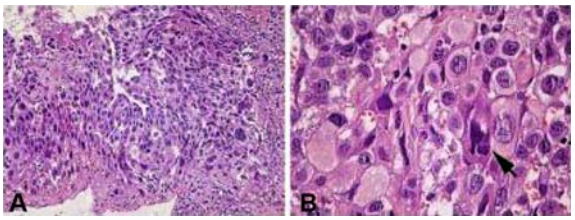

Fig. 3 Histological findings
A) A low power field histologic figure of H&E stained, there is a sheet malignant epithelial cells shows marked nuclear pleomorphism
B) A high power field shows syncytiotrophiblast-like multinucleated giant cells with pyknotic nuclei (arrow) are occasionally seen uninucleated malignant cells

Discussion

Primary choriocarcinoma of the uterine cervix is an extremely rare disease. Saito et al first described the diagnostic criteria of such an entity, that were, (1) absence of disease in uterine cavity, (2) pathologic confirmation of disease, (3) exclusion of molar pregnancy, and (4) exclusion of coexisting normal intrauterine pregnancy[2]. Because of low suspicion due to its rarity and the majority of cases presented with abnormal vaginal bleeding which could be caused by

other more common conditions including, threatened abortion, cervical polyp, cervical pregnancy, and cervical cancer. Therefore, the accurate diagnosis of primary cervical choriocarcinoma is difficult to achieve[1,3-8]. In the presented case, the origin of choriocarcinoma is still inconclusive. Possibly, it would be a consequence of the last abortion. In the presented case, the patient came with massive vaginal bleeding. The primary cervical choriocarcinoma was misdiagnosed as a bulky cervical cancer and multiple punch biopsies were further performed with unawareness of life-threatening vaginal bleeding. Despite its rarity, cervical choriocarcinoma should therefore, be considered in differential diagnosis of patients who present with cervical mass and profuse bleeding particularly in young women and cervical biopsy should be avoided in cases with highly suspicion. The initial pathologic report of biopsy specimens in the presented case was also misdiagnosed as poorly differentiated squamous cell carcinoma. The difficulty for the pathologist to distinguish cervical squamous cell carcinoma particularly poorly differentiated type from choriocarcinoma has been well described[1,3,5,6]. The detection of serum beta-hCG level and the pathologic slide review confirmed by immunocytochemistry staining were helpful in the diagnosis as presented in the present report.

In the literature, almost all cases with cervical choriocarcinoma were initially treated with total hysterectomy due to uncontrollable bleeding[1,3,5,6]. With the advance of radio-intervention techniques, the present case highlighted the success of selective uterine arterial embolization to control profuse bleeding from cervical choriocarcinoma.

Despite the growing progression of chemotherapy in GTD which lets almost all patients be cured with chemotherapy alone, the reported experience of the use of chemotherapy without hysterectomy for cervical choriocarcinoma was limited[6]. The successful treatment of cervical choriocarcinoma by chemotherapy alone in the present case, therefore, offers the awareness of the possible chance of conservative management for cervical choriocarcinoma even in cases with a large lesion.

In conclusion, cervical choriocarcinoma should be considered in the differential diagnosis of a cervical mass with profuse bleeding particularly in young women. Selective uterine arterial embolization may be used to stop bleeding in patients who have hemodynamically stable condition and wish to maintain future fertility.

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การรักษาแบบประคับประคองในมะเร็งเนื้อรกขนาดใหญ่ของปากมดลูกที่มีเลือดออกรุนแรง

อัญชลี จันทร์แจ่ม, ชำนาญ เกียรติพีรกุล, สุรพันธ์ คุณอมรพงศ์, ประภาพร สู่ประเสริฐ, จตุพล ศรีสมบูรณ์, ชัยเลิศ พงษ์นริศร, ฉลอง ชีวเกรียงไกร, สิทธิชา สิริอารีย์, จารุวรรณ ตันติพลากร

มะเร็งเนื้อรกของปากมดลูกนั้นพบได้ค่อนข้างน้อยเนื่องจากเป็นโรคที่นึกถึงน้อย และผู้ป่วยส่วนใหญ่จะมาด้วยอาการเลือดออกผิดปกติทางช่องคลอดซึ่งอาจเกิดจากภาวะอื่นได้บ่อยกว่า เช่น ภาวะแท้งบุตร, ติ่งเนื้อ ที่ปากมดลูก, การตั้งครรภ์ที่ปากมดลูก หรือ มะเร็งปากมดลูก การวินิจฉัยที่แน่นอนของโรคนี้จึงค่อนข้างยาก ในรายงานนี้ผู้ป่วยเป็นมะเร็งเนื้อรกของปากมดลูกที่มีขนาดใหญ่และมีเลือดออกทางช่องคลอดปริมาณมาก โดยในตอนแรกได้รับการวินิจฉัยว่าเป็นมะเร็งปากมดลูก เลือดที่ออกมากจากปากมดลูกสามารถควบคุมได้โดยการทำให้หลอดเลือดแดงที่ไปเลี้ยงมดลูกอุดตัน และใช้การรักษาด้วยยาเคมีบำบัดแบบยารวมทำให้มีระยะสงบของโรคได้โดยไม่ต้องผ่าตัดเอามดลูกออก