

## Case Report

# Spontaneous rupture of cervical hydatidiform molar pregnancy in 53 years old woman: A case report and brief review of the literature

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### Abstract

The incidence of molar pregnancy and ectopic pregnancy are 1 and 10 per 1000 pregnancies, respectively. An ectopic molar pregnancy is a very rare occurrence. The differentiation of molar ectopic and non-molar ectopic pregnancy is very difficult. A 53 years old gravida 5, parity 2, dilatation and curettage 1, abortus 1 woman was referred from emergency service due to excessive vaginal bleeding and pelvic pain. Pelvic examination revealed significant vaginal hemorrhage from a lesion on anterior cervical lip. Quantitative  $\beta$ -hCG test was 41.842mU/ml. Ultrasonographic findings revealed no bilateral adnexal or uterine pathology but cervical ectopic pregnancy. As the patient's hemoglobin level was 4.7 gr/dL, total abdominal hysterectomy and bilateral salpingo-oophorectomy was performed immediately. Final pathologic examination revealed cervical hydatidiform molar pregnancy. No peri or postoperative complication was detected and the patient was uneventful.

### Key words:

Pregnancy, cervical ectopic pregnancy, uterine cervix, hydatidiform mole

## Introduction

Ectopic pregnancy occurs when the developing blastocyst becomes implanted at a site other than the endometrium of the uterine cavity. The prevalence of ectopic pregnancy among women who go to an emergency department with first trimester bleeding, pain, or both ranges from 6 to 16 % [1]. However, the overall incidence of ectopic pregnancy increased during the mid-twentieth century, plateauing at approximately almost 20 per 1000 pregnancies in the early 1990s, the recent national data were reported by the Centers for Disease Control [2]. Previous ectopic pregnancy, tubal pathology and surgery, in-utero DES exposure, previous genital infections, intrauterine devices, infertility, multiple sexual partners, smoking, in vitro fertilization, vaginal douching, age are known risk factors for ectopic pregnancy.

Cervical pregnancy is a rare form of ectopic pregnancy in which the trophoblast implants in the cervical tissue of the endocervical canal. Cervical pregnancy incidence is approximately 1/9000 [3,4]. Although the etiology is still unknown, there is evidence for its association with

cervico-uterine instrumentation [3], and in particular vaginal termination of pregnancy [5]. Several case reports have suggested that invitro fertilization may also increase the risk of cervical ectopic pregnancy [6,7].

Gestational trophoblastic disease is a proliferative disorder of trophoblastic cells. It defines a heterogeneous group of interrelated lesions arising from the trophoblastic epithelium of the placenta. The World Health Organization classification of gestational trophoblastic disease includes complete and partial hydatidiform mole, invasive mole, chorioncarcinoma, placental site trophoblastic tumor, epitheloid trophoblastic tumor, exaggerated placental site and placental site nodule [8].

Hydatidiform molar degeneration of the placenta tissue can be classified in partial and complete hydatidiform mole pregnancy. Hydatidiform mole pregnancy is considered as a sporadic and genetic disorder with a recurrence rate of 1%. Risk factors for the development of hydatidiform molar pregnancy are maternal age, history of previous molar pregnancy, smoking, alcohol abuse, oral contraceptives [9,10]. Normally molar pregnancy presents in the first trimester and is associated with a wide array of clinical symptoms, most commonly vaginal bleeding in combination with excessive  $\beta$ -HCG levels. However at the time of presentation, 60 % of molar gestation had ruptured [11].

Cervical hydatidiform molar pregnancy is not common and only four case reports are presented in the literature [12-15]. Here is a presentation one more case of cervical molar pregnancy in a 53 year old woman.

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## Case presentation

A 53 years old gravida 5 parity 2 dilatation and curettage 1, abortus 1 woman was admitted to our emergency department with complaint of vaginal bleeding for 15 days and pelvic pain. Pelvic examination revealed significant vaginal hemorrhage from ruptured lesion on cervical lip. Ultrasonographic findings revealed no bilateral adnexal or uterine pathology but cervical ectopic pregnancy. Materials in the ruptured lesion on cervical lip were removed for pathologic examination and gentle aspiration curettage was performed to cease bleeding. As the patient's quantitative  $\beta$ -hCG test was 41.842 mU/ml and hemoglobin level were 4.7 gr/dL, total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. On gross examination 2x1 cm area on the serosal surface of left side of cervix was ruptured (Figure 1). Microscopic evaluation revealed endocervical glands and stroma consisting of large cells with eosinophilic cytoplasm with hyperchromatic nuclei (Figure 2). Immunohistochemically, these cells expressed cytokeratin AE1-AE3 but not vimentin (Figure 3). The findings supported that these cells were trophoblastic in nature. Histologically, materials derived from ruptured cervical lesion showed cisternal formations in the stromal parts of the chorionic villi. Moreover, there were significant abnormal proliferations in the trophoblastic cells with areas displaying circumferential growth from the villous surface (Figure 4). Five weeks after the operation,  $\beta$ -HCG level was 4.7mU/ml and the patient was uneventful.

**Figure 1.**

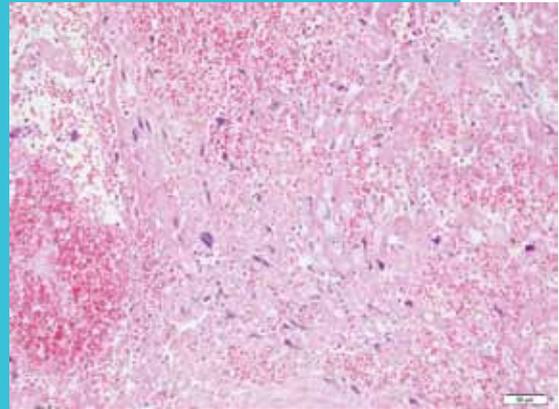


*Figure 1 Macroscopic appearance of ruptured serosal surface of left side of uterine cervix*

## Discussion

Incidence of cervical ectopic pregnancy is approximately 1/9000 [3, 4]. It may be fatal due to excessive vaginal bleeding; fortunately, early diagnosis and effective treatment methods significantly decrease the mortality rates. No recent deaths due to cervical pregnancy have been reported [1]. Ectopic molar pregnancy is an extremely rare condition. The incidence of ectopic gestational trophoblastic disease can be estimated at approximately 1.5 per 1000000 births [11].

**Figure 2.**



*Figure 2 Endocervical glands and stroma consisting of large cells with eosinophilic cytoplasm with hyperchromatic nuclei, microscopically (H&Ex10)*

To the best of our knowledge, this is the fifth case of cervical molar pregnancy in PUBMED as shown in Table 1 [12-15]. Two of these cases were partial molar pregnancy [12, 14]. In these two cases, dilatation and curettage was performed and bleeding ended after operation. The third case was a complete hydatiform molar pregnancy of the cervix presenting with vaginal bleeding. In this case methotrexate treatment was attempted first, but heavy hemorrhage necessitated evacuation. Moreover, patient had demonstrated raised  $\beta$ -HCG levels that remained elevated for up to 4 weeks, was given a chemotherapy treatment consisting of seven successive doses of methotrexate given in combination with folinic acid [13]. Fourth case was complete molar ectopic pregnancy localized to the ectocervix. This case occurred two months after curettage of a missed abortion [15]. As the patient in the present case was hemodynamically unstable and in menopausal transition period, total abdominal hysterectomy and bilateral salpingo-oophorectomy was performed.

In conclusion, physicians should keep in mind that, cervical ectopic molar pregnancy may cause excessive vaginal bleeding in a woman even at her fifth decade.

Figure 3.

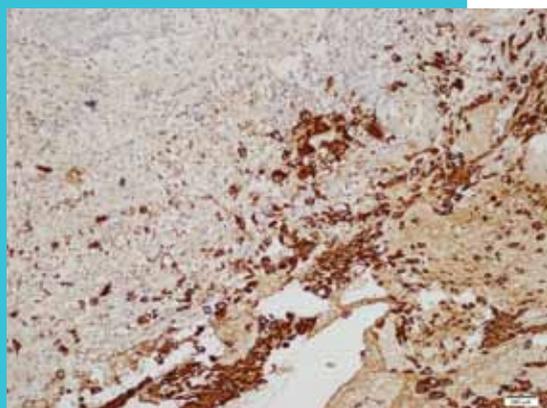


Figure 3 Cells expressing cytokeratin AE1-AE3 but not vimentin, immunohistochemically

Figure 4.

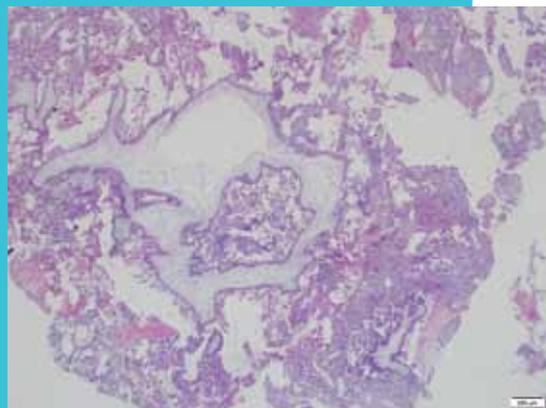


Figure 4 Abnormal proliferations in the trophoblastic cells with areas displaying circumferential growth from the villous surface (H&Ex10)

#### Conflict of interest statement

The authors declare no conflict of interest.

Table 1 Comparison of the present case and the previous literature regarding cervical ectopic molar pregnancy

Reference	Age	Clinical Presentation	Histology	Treatment	D Follow-up
K Chapman [12]	35	Elevated vaginal bleeding for the last 2 days that has been occurring for the past 3 weeks, lower abdominal pain	Partial mole	D&C	After 6 weeks, $\beta$ -hCG <5U/L
Wee et al., [13]	36	Heavy vaginal bleeding following the evacuation done 2 weeks ago	Complete mole	Mtx, evacuation, and bimanual compression. Afterwards, chemotherapy with mtx, 7 cure.	14 weeks later, $\beta$ -hCG <2IU/L
Aytan et al., [14]	25	Heavy vaginal bleeding	Partial mole	D&C, bimanual compression	3 weeks later, $\beta$ -hCG <5mIU/ml
Schwentner et al., [15]	28	Heavy vaginal bleeding following the abortion done 2 months ago	Complete mole	Exfoliation and conservative surgery	After 3 months, beta- $\beta$ -hCG undetectable.
Present case	53	Heavy vaginal bleeding	Complete mole	TAH+BSO	After 5 weeks, $\beta$ -hCG <5mIU/ml

D&C= Dilatation and curettage, Mtx= Methotrexate, TAH+BSO= total abdominal hysterectomy and bilateral salpingo-oophorectomy

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